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Foreign Accent Syndrome: A Neurolinguistic Analysis

Keulen, Stefanie Agnes Marino Theo

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Foreign Accent Syndrome: A Neurolinguistic Analysis

STEFANIE KEULEN

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The work reported in this thesis has been carried out under the auspices of the Center for Language and Cognition Groningen (CLCG) at the University of Groningen (NL) and the Center for Linguistics at the Vrije Universiteit Brussel (BE).
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VRIJE UNIVERSITEIT BRUSSEL

Foreign Accent Syndrome: A Neurolinguistic Analysis

PhD thesis

to obtain the degree of PhD of the University of Groningen on the authority of the Rector Magnificus Prof. E. Sterken, and in accordance with the decision by the College of Deans

and

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by

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LIST OF ABBREVIATIONS

ABA-2 - Apraxia Battery for Adults-2
ADAS - Alzheimer's Disease Assessment Scale
ADL - Activities of Daily Living
AIDS - Assessment of Intelligibility of Dysarthric Speech
AoS - Apraxia of Speech
APA - American Psychiatric Association
AQ - Aphasia Quotient
ASHA - American Speech-Language-Hearing Association
BA - Brodmann Area
BADA - Batteria per l'analisi dei deficit afasici
BDAE - Boston Diagnostic Aphasia Examination
BDI - Beck Depression Inventory
BNT - Boston Naming Test
BVMT-R - Brief Visuospatial Memory Test - Revised
CAS - Childhood Apraxia of Speech
CCAS - Cerebellar Cognitive Affective Syndrome
CEFR- Common European Framework of Reference for Languages
CES-D - Center for Epidemiologic Studies-Depression Scale
CLQT - Cognitive Linguistic Quick Test
COWAT - Controlled Oral Word Association Task
CVLT - California Verbal Learning Test
CT - Computed Tomography
d - day(s)
DAS - Developmental Apraxia of Speech
DBS - Deep Brain Stimulation
DES - Direct Electrical Stimulation
D-KEFS - Delis-Kaplan Executive Function Scale
DISQ-R - Dissociation Questionnaire -Revised
DO-80 - Test de Dénomination Orale d'Images
DSM - Diagnostic and Statistical Manual of Mental Disorders
DSQ-60 - Defense Style Questionnaire - 60
DTI - Diffusion Tensor Imaging
DWI - Diffusion Weighted Imaging
EEG - Electroencephalogram
EPVS - Expanded Perivascular Spaces
F - Female
FAS - Foreign Accent Syndrome
FSIQ - Full Scale Intelligence Quotient
HADS - Hospital Anxiety and Depression Scale
HBV - Hepatitis B Virus

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HCV - Hepatitis C Virus
HDS - Hierarchic Dementia Scale
HIV - Human Immunodeficiency Virus
IQ - Intelligence Quotient
ICC - Intraclass Correlation Coefficient
K-FAST - Korean version of Frenchay Aphasia Screening
L1 - First language
L2 - Second language
L3 - Third language
LIPP - List of Indiscriminate Psychopathology
m - Month(s)
M - Male/Mean (depending on chapter)
MAE - Multilingual Aphasic Examination
Max - Maximum
MCA - Middle Cerebral Artery
Min - Minimum
MMPI - Minnesota Multiphasic Personality Inventory
MMSE - Mini Mental State Examination
MRI - Magnetic Resonance Imaging
MS - Multiple Sclerosis
MVA - Motor Vehicle Accident
NA - Not Administered
NEO-PI-R - Revised Neuroticism-Extraversion-Openness Personality Inventory
NPI-40 - Narcissistic Personality Inventory - 40
OCD - Obsessive-compulsive Disorder
PALPA - Psycholinguistic Assessments of Language Processing in Aphasia
pc. - Percentile
PENO - Protocole d'Evaluation Neuropsychologique Optimal
PFC - Prefrontal Cortex
PIQ - Performance Intelligence Quotient
PNI - Pathological Narcissism Inventory
PTSD - Post-Traumatic Stress Disorder
PPTT - Pyramid and Palm Tree Test
PVI - Pairwise Variability Index
PVWM - Periventricular White Matter
RAVLT - Rey Auditory Verbal Learning Test
RCF - Rey Complex Figure
RCPM - Raven Colored Progressive Matrices
RISB - FR - Rotter Incomplete Sentences Blank - French version
ROCF - Rey-Osterrieth Complex Figure Test
ROI - Region of Interest
SAN-Test - Stichting Afasia Nederland - Test

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SCL-90-R - Symptom Checklist-90-Revised
SD - Standard Deviation
SLI - Specific Language Impairment
SPECT - Single Positron Emission Computed Tomography
SS - Supernormality Scale
STAI - State-Trait Anxiety Inventory
STG - Superior Temporal Gyrus
TIA - Transient Ischemic Attack
TMT - Trail Making Test
VIQ - Verbal Intelligence Quotient
WAB - Western Aphasia Battery
WAIS - Wechsler Adult Intelligence Scale
WCST - Wisconsin Card Sorting Test
WM - White Matter
WMS - Wechsler Memory Scale
WMS-R - Wechsler Memory Scale-Revised
WRAT - Wide Range of Achievement Test
y - year(s)

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CHAPTER 1

General Introduction

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The aim of this dissertation is to provide a neurolinguistic description of the speech disorder called foreign accent syndrome (FAS). FAS is a rare motor speech impairment that affects the patients in such a way that they start speaking their native language with a foreign accent. Verhoeven & Mariën (2010a) have identified three major subtypes of foreign accent syndrome: a neurogenic type (including a developmental one), a psychogenic type and a mixed type. This dissertation aims at providing the reader with (1) an in-depth analysis of the neurolinguistic features associated with neurogenic, developmental and psychogenic FAS, (2) an analysis of the pathophysiological mechanism responsible for this change of accent and (3) an analysis of the position of FAS within the spectrum of motor speech disorders. The objectives will be realized via a systematic research synthesis (of published literature and case studies), as well as a series of new case reports. Below we first present a brief history (1.1) of the conceptualization and evolution of the disorder and discuss its operationalization by Whitaker (1982) (1.2). Subsequently we address the taxonomic distinction made by Verhoeven & Mariën (2010a) (1.3). Afterwards we provide an overview of the most influential theoretical accounts on FAS (1.4).

1.1. YOUNG AND MISUNDERSTOOD: FOREIGN ACCENT SYNDROME IN THE EARLY DAYS

1.1.1. Brief historical overview of the motor speech disorders

Motor speech disorders are among the most explicit signs of neurological damage. Speech is such a profoundly human, though complex cognitive-motor function, that a breakdown of its mechanisms is considered most invalidating. This dissertation does not focus on the breakdown of language and communication, but rather on impairments of speech or: the production of sound waves to convey a message. According to Prins & Bastiaanse (2006) the earliest references to speechlessness after head trauma date back to the so-called Edwin Smith surgical papyrus. It is believed to have been written around approximately 1700 BC. They also speak of a Hittite text elaborating on the possible aphasia (?) incurred by King Mursilis in 1400 BC. Next, Prins & Bastiaanse (2006), Weismer (2007), and Freed (2012) all speak of the references to motor speech and/or language disorders in the works of Greek philosopher Hippocrates (460-370 BC).

In 'Aphorisms' (400 BC) and 'Of the Epidemics' (400 BC) Hippocrates describes patients in whom there was a sudden loss of speech. However, in these instances it is not quite clear whether the patient suffered aphasia or a motor speech disorder. Advances in neuroanatomy were crucial to the clearer description of the more well-known motor speech disorders of today. Two of them that are quite crucial in description of foreign accent syndrome: namely apraxia of speech and dysarthria.

The first clear account of dysarthria possibly dates back to the 1600s and has been attributed to Thomas Willis who delivered some ground-breaking works in neuroana-

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tomy, such as the description of the circle of Willis (Willis, 1664). He described the branches of the nervus vagus and the spinal nerves. Willis also gave an account of the speech patterns of a female patient who suffered myasthenia gravis, which was later argued to be an instance of (flaccid) dysarthria (Weismer, 2007). In 1817, James Parkinson (1755-1824) noted anomalies in the speech of patients with so-called shaking palsy (later denoted patients with 'Parkinson's Disease'), which would be identified as a hypokinetic dysarthria. Ataxic dysarthria was possibly reported for the first time by Jean Martin Charcot (1825-1893) in association with multiple sclerosis (MS). In 1868 he argued that cerebellar dysarthria occurs in MS when the triad of Charcot is diagnosed (nystagmus, intention tremor, scanned speech) (Schönle & Gröne, 1993). However, today these patients are more commonly diagnosed with mixed dysarthria; which is the common denominator for a series of characteristics typifying several types of dysarthria at once. In 1917, Gordon Holmes (1876–1965) described a motor speech disorder in association to cerebellar pathology. The characteristics he enumerates in his description still correspond well to the description of ataxic dysarthria by Darley et al. (1969b) (Mariën & Beaton, 2014; Ziegler, 2016). Although the first account of hyperkinetic dysarthria is not clear, the disorder has been commonly associated with Huntington's disease. In 1872, Georges Huntington's account 'on chorea' was published, and although he did not recognize in these 'Huntington patients' a true speech disorder, he speaks of Romberg's account of two patients whose respiration was affected. Another form of hyperkinetic dysarthria, the dystonic type, is now commonly associated with 'Wilson's disease', which was described by Kinnier Wilson in 1912. The formal taxonomy of dysarthria, including the diagnostic features, was only established in the 20th century, by Darley et al. (1969b, 1975) and included flaccid, spastic, hypokinetic, hyperkinetic and ataxic dysarthria as well as the mixed type dysarthria. Although 1876, Marie & Katwinkel supposedly gave the first account of unilateral upper motor neuron dysarthria (Freed, 2012). Darley et al. did not include this variant of dysarthria in their overview (1969b; 1997, see also Darley 1969). According to Duffy (2013) this is because most of the research on the consequences of upper motor neuron lesions on speech only started as of the 1980's.

In 1900, Hugo Karl Liepmann (1863-1925) gave the first account of 'apraxia', and distinguished between ideomotor, limb-kinetic or innervatory, and ideational apraxia. He equally reported the 'apraxia of the glosso-labio-pharyngeal structures'. An apraxia causes a failure to execute voluntary acts when muscle strength is preserved. This clearly distinguishes the disorder from dysarthria, in which there is impairment at the muscular level.

The term 'apraxia of speech' was coined by Darley in 1969 (Ogar et al., 2005). Problematic is that the current concept of apraxia of speech has had a very elaborate evolution with regards to its terminology (to the frustration of many researchers...). The first reference is attributed to Broca (1861), who called it 'aphemia' and stated (with respect to his famous patient, Mr. Leborgne):

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"There are cases in which the general faculty for language remains unaltered; where the auditory apparatus is intact; where all muscles – including those of speech and articulation – are under voluntary control; and where nevertheless, a cerebral lesion abolishes articulated language." (Broca, 1861)

Trousseau (1864) increased the confusion by referring to 'aphasia'; when he was describing features essential to today's apraxia of speech (Tesak & Code, 2008). John Hughlings Jackson (1864) described today's apraxia of speech as 'speechlessness', and gave an account that was informative to many others to follow, including Marie (1906), who would refer to it as 'anarthria' after reviewing the accounts of amongst others, Trousseau (1864) and Carl Wernicke (1874).

Jackson (1864) described an instance of today's 'aphemia' in his patient, and especially pointed out the remarkable automatic-voluntary dissociation:

"In some cases of defective speech the patient seems to have lost much of his power to do anything he is told to do even with those muscles that are not paralyzed. Thus a patient will be unable to put out his tongue when asked him although he will use it well in semi-voluntary actions, e.g. eating and swallowing. He will not make a grimace he is told to do, even when we make one for him to imitate. There is power in his muscles and in the centers for the coordination of muscular groups but he, the whole man or the "will" cannot set him agoing. Such a patient may do a thing well at one time but not another." (cited in Head, 1915, pp. 36-37).

He furthermore states that:

"The words of the speechless patient are not at his disposal for voluntary use; they exist for comprehension and can also be called upon under emotional stress but, like the movement of the tongue, they cannot be reached by the will" (cited in Head, 1926, p. 36)

Interestingly, until Liepmann's description, only Hughlings Jackson regarded the disorder as situated at the level of speech; whereas all the other scientists saw apraxia of speech as a *language* disorder. 1923 was another important landmark date in the conceptualization of apraxia of speech. Alajouanine, Ombredane & Durand (1939) published their description of the 'phonetic disintegration syndrome' in aphasic patients suffering from anterior lesions. Specific segmental alterations were pointed out, including different consonant substitutions and vowel substitutions. Interestingly however, they believed the changes were not always strictly due to articulatory disturbances (at the planning level) but equally left room for interference at the muscular level (changes due to paresis, dystonia, ...) (Micoch & Square, 1984; Denes & Pizzamiglio 1999). In 1969, apraxia of speech was unequivocally defined and its characteristics were enumerated by Darley (1969). Although confusion continued up until the seventies, as 'apraxia of speech' also coincided with Luria's notion of 'afferent motor aphasia' (1970), for instance.

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FAS was – to our knowledge – only first described in the early 20th century. In 1907, Pierre Marie (1853-1940) presented an account of a Parisian man who incurred a hemorrhage in the quadrilateral area of Marie, the lenticular zone of the left-hemisphere (i.e. the first temporal convolution). The patient was hemiplegic and developed a pure anarthria after recovery from mutism. Marie's anarthria is now commonly termed an apraxia of speech. The patient's mutism remained present for nine months. When the patient regained his speech, he started speaking with an Alsatian accent. This first anecdotal report represents a change to a regional language. Marie clearly regards the FAS as a remnant of the initial apraxia of speech.

1.1.2. From Pierre Marie onwards...

The next case was published only 12 years later (1919), by Arnold Pick (1851-1924). He described FAS in a 26-year-old Czech butcher. His patient suffered a left-hemisphere stroke that left him with a non-fluent aphasia, agrammatism, writing deficits (the patient could only write his name), and a remarkable accent change (Polish accent). Interestingly however, the patient had been stationed in Poland as a soldier for several months during the war. Hence, this might have been an account of a polyglot or bilingual aphasia; i.e. an aphasia in a bilingual or polyglot patient affecting all of the languages the patient masters (Fabbro, 2001). The aphasia can also be differential: i.e. affecting different languages to a different degree (Albert & Obler, 1978). Pick's description of his patient is important because he connects the accent change to a series of segmental and suprasegmental changes which he described in some detail: Pick reported the Polish sounding-accent was due to a change in word accent (stressing the antepenultimate syllable, instead of the first one, as is expected in Czech), a shortening of vowels, a softening of fricatives and affricates, and nasalizing vowels. Pick argues that the change of accent was the result of a wrong word accent placement and attenuation of fricatives.

The following case was reported in 1947 by the Norwegian neurologist Georg Monrad-Krohn (1884-1964). Monrad-Krohn was the first to report the psychosocial implications of an accent change. He presented the case of the 30-year-old Norwegian woman, Astrid L., who incurred a hemorrhage affecting a vast area of the left fronto-temporo-parietal region after being hit by bomb shrapnel during an air raid (WW II). For the first time, a radiograph was included in the analysis and the lesion location was described in some detail. Initial neurological symptoms were right hemiplegia and agrammatic aphasia. Two years post-injury, when Monrad-Krohn saw his patient for the first time, she was still mildly aphasic and dysgraphic and she spoke with a clear German accent. The patient suffered major social exclusion during the Second World War as her German accent was very poorly viewed in an occupied country. Monrad-Krohn saw a connection between Astrid L. and the patient described by Arnold Pick: both had undergone comparable changes in their speech. However, whereas Pick attributed the change to segmental and prosodic changes, for Monrad-Krohn they were

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largely due to suprasegmental deficits. Norwegian has two distinct pitch patterns to differentiate (polysyllabic) homonyms: a 'one-syllable-accent' and a 'two-syllable-accent'. The patient had difficulties applying these accents correctly. For Monrad-Krohn, the attainment of the patient's speech had to consist of a prosodic impairment and he coined the term 'dysprosody'.

In 1961, Nielsen & McKeown reported two English-speaking American patients who developed a Swedish accent after a thrombosis (case 1) and brain contusions (case 2). They contested Monrad-Krohn's vision that FAS was a purely prosodic disorder: it was likely a motor speech disorder resembling dysarthria. The authors emphasize the subjectiveness in the diagnosis of a prosodic disturbance:

"...the dysprosody is simply interpreted as dysarthria unless it reminds the listener of some particular foreign language, and that therefore the interpretation is largely subjective. If the examiner did not have acquaintance with any language but his own, he would not see a simulation of some foreign language but only a peculiar form of dysarthria, a disturbance of rhythm" (p. 159).

In 1962, Macdonald Critchley (1900-1997) gave the account of three patients who experienced a change of regional accent and a reversion to a previously learned accent. He distinguishes between dysphonemia and dysprosodia, which could be interpreted as disorders affecting segmental and suprasegmental characteristics respectively. The first patient was a 49-year-old woman who suffered a post-traumatic stress disorder after being involved in a car crash. The possibility of structural brain damage is not discussed. Critchley argued that the case was "almost certainly psychologically determined" (p. 185). The patient's only symptom was that she started speaking with what was perceived to be a Welsh accent after the crash. The second case was a 48-year-old lady who had suffered a stroke causing her to speak with a French accent. The patient was raised bilingually in French and English, although she had not used French for a long time when the event occurred. The third patient was a 37-year-old right-handed woman from the UK who incurred a stroke, resulting in a left hemiplegia and mutism. The patient had expressive and comprehensive deficits (reading) and suffered from aphasia. She gradually regained speech, although she started speaking with a marked Welsh accent. Interestingly, her family stated the patient had undergone a personality change after suffering her stroke. The patient disliked speaking with a regional accent, possibly due to the association with a failed marriage with a Welshman. If lesion location would have been formally confirmed, this may have constituted the first instance of crossed FAS. According to Critchley, latter case is exemplary for the post-aphasic dysprosodia in the way Monrad-Krohn (1946) described it, although Critchley himself argued to distinguish distortions at the level of phonemes ('dysphonemia') as well.

Two years later, Charles William Michael Whitty (1914-1996) described a 27-year-old, right-handed patient who got surgery for an angioma in the left MCA territory. In the acute stage the patient was mute, suffered orofacial apraxia and a cortical dysarthria

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(i.e. an apraxia of speech). The orofacial apraxia and muteness ("complete anarthria", p. 509) receded within 36 hours post-onset. The patient continued to suffer from articulatory problems (with an automatic-voluntary dissociation, see also above), an apraxic dysgraphia, and speech had undergone a change in accent: although the patient was a monolingual speaker of English (with some basic knowledge of French), friends and relatives believed she spoke with a German accent. Interestingly also, Whitty argued symptoms were related in the sense that when apraxic speech was more audible, the accent increased as well. Whitty on the one hand argues that segmental analysis may demonstrate that it is "part of a spectrum of basically executive alterations of speech", and just after that "[f]or the present it must be noted as an unusual and bizarre aspect of motor dysphasia [i.e. a non-fluent aphasia, *our comment*]" (p. 510), a remark which is in line with the theories of Monrad-Krohn (1946) and Critchley (1962), who agree that the change is some kind of post-aphasic dysprosodia. Interestingly, craniotomy showed that the lesion was actually situated within Marie's lenticular area, which comprises the insula, and could explain the apraxic symptoms. This indicates that the accent change could have been of an apraxic nature as well. The accent remained present for 7 weeks post-onset. According to Whitty, it was the intact right-hemisphere that subserved a crucial role in the repair of speech and melody.

In 1971, Monroe Cole (1933-2008) published a particularly interesting paper including two new cases of sudden accent change. The first case was a 29-year-old man who had suffered a cerebellar degeneration after anoxia. After the event, he displayed neurological symptoms that included spastic-ataxic gait, bilateral extensor-plantar response and ataxia of arms and legs. Speech was qualified as slurred and dysarthric and although the patient was from Ohio and spoke both American English and Yiddish premorbidly, his family argued that his speech now sounded Italian. The patient had studied Italian, although he stated he did not master this language anymore.

The second case was a 58-year-old woman who developed similar neurological symptoms: bilateral extensor plantar response, ataxic gait, vertical and horizontal nystagmus and dysarthric speech. The patient was diagnosed with a pontine infarction. Relatives argued that after the stroke, the patient's accent changed from Ohian to 'Eastern European', or Hungarian-like.

In the article, Cole argues that there is a close relationship between the cortical dysarthria, or apraxia of speech, and 'posterior fossa dysarthria' or ataxic dysarthria. Cole also emphasized that both his patients had been exposed to the language their newly acquired accent was associated with, although they did not – or no longer – speak these languages.

Cole does not discuss the role of the cerebellum in the onset of the accent change, or prosodic disturbance, even though he notices an association with ataxic dysarthria.

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1.2. THE DEVELOPMENT OF FAS CRITERIA

The descriptions of the disorder – which still had no name – remain speculative. In 1982, Harry Whitaker publishes his seminal paper and coins the term 'foreign accent syndrome'. He defines the criteria for its diagnosis, based on three case studies: the ones by Pick (1919) and Monrad-Krohn (1947), chosen because of their completeness with regards to the anatomo-behavioral data as well as the linguistic descriptions, and his own, new case. He defines the FAS-criteria as follows:

"1. The accent is considered by the patient, by acquaintances and by the investigator to sound foreign
2. it is unlike the patient's native dialect before the cerebral insult
3. it is clearly related to central nervous system damage (as opposed to an hysteric reaction, if such exist)
4. and there is no evidence in the patient's background of being a speaker of a foreign language (i.e., this is not like cases of polyglot aphasia)" (Whitaker, 1982, p. 196,198)

Only Pick's patient might actually have represented an instance of polyglot aphasia. The patient had resided for several months in Poland during the war. Moreover, many of the other, previously mentioned instances of accent change described before could not be diagnosed as 'FAS' based on this account, as they presented a shift to a (regional) accent that the patient definitely was familiar with (e.g. Critchley 1962, case 1, 3) or there was a change in a bilingual patient (Critchley, case 2) or the patient at least had learned and/or was fluent in another language at some point in his/her life (Whitty, 1964). What then to do with these cases?

Whitaker's patient was a 30-year-old right-handed American woman, who arrived at the hospital with complaints of an accent change. A few months earlier, she had suffered a stroke that had left her with an aphasia with agrammatism and an apraxia of speech. After resolution of these symptoms, a Spanish-sounding accent remained. Interestingly, the patient had been in contact with Spanish-speaking people, although she could not speak the language herself. Neurological origin of the symptoms was not confirmed via neuroimaging. Whitaker argues that the lesion site must have been situated in the territory of the left MCA and may have encroached upon Broca's area. Whitaker summarized the most poignant changes in his patient's speech as (1) changes against the place of articulation of consonants, (2) consonant omission, (3) consonant substitution, (4) consonant devoicing, (5) monophthongization, and (6) vowel fronting and raising. Interestingly, Whitaker argued, based on a personal account of the patient in conjunction with the findings at hand, that the timing errors were analogous to what is encountered in ataxic dysarthria. The patient seemed unable to reach speech targets in voluntary speech control. He also argued that – based on the cases by Cole (1971) – cerebellar functioning could have been disturbed as this structure sends projections to the area of Broca.

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The majority of the cerebral input to cerebellum has been proven to originate from the premotor and primary motor cortices (Purves et al., 2001), and the cerebellum similarly sends output to these regions (Schmahmann & Pandya, 1997; Dum & Strick, 2002). A relation between FAS and the cerebellum thus seems less speculative than Whitaker thought.

1.3. A TAXONOMIC DISTINCTION

After the formulation of the FAS criteria, many cases of 'foreign accent syndrome' were published that did not fully comply with the criteria proposed by Whitaker. Between 1982 and 2010, approximately 63 original case reports were published in the literature. However, at least six patients did not have CNS damage and demonstrated a psychiatric disorder (Reeves & Norton, 2001; Van Borsel et al., 2005; Verhoeven et al., 2005; Reeves et al., 2007, case 1 & 2; Tsuruga et al., 2008). In six other patients, CNS damage could not be confirmed and the etiology remained speculative (Gurd et al., 2001; Hwang et al., 2001; Laures-Gore et al., 2006; Moen 2006; Poulin et al., 2007, Katz et al., 2008). Moreover, ten patients violated the criterion concerning linguistic background, as they were bilinguals or polyglots (Schiff et al., 1983; Seliger et al., 1992; Kurowski et al., 1996; Roth et al., 1997; Verhoeven & Mariën, 2002; Avila et al., 2004; Lippert-Gruener et al., 2005; Verhoeven et al., 2005; Paquier & Assal, 2007, Chanson et al., 2009, Mariën et al., 2009).

That is why, in 2010, Verhoeven & Mariën published a taxomic overview of FAS. They distinguished between three different types: a neurogenic type, a psychogenic type and a mixed FAS. The neurogenic FAS, which corresponds most to Whitaker's FAS' description, can be further subdivided in an acquired and a developmental type: in the acquired type, FAS develops after neurological damage of an acquired nature (e.g. stroke), whereas developmental FAS affects language as of development. In the psychogenic type, FAS is related to a psychiatric disorder. The mixed subtype – as the term already hints – is a FAS acquired on a neurological basis. However, the patient develops a new identity in order to achieve a better correspondence between the newly acquired accent and his/her personality (he/she undergoes a psychological change of some sort).

1.4. PERSPECTIVES ON FOREIGN ACCENT SYNDROME

Although Verhoeven & Mariën's (2010a) paper was groundbreaking in the sense that it defined the subtypes, researchers continued to look for explanations as to what gave rise to the accent change. Till 2010 these accounts were mainly linguistically (see 1.4.1.; e.g. Ingram et al., 1992) or cognitively based (1.4.2.; e.g. Whiteside & Varley, 1998), whereas afterwards, computational models were introduced (e.g. 1.4.3.; Tomasino et

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al., 2013). Interestingly, most of these were aimed at explaining neurogenic FAS and the remaining subtypes were not discussed.

1.4.1. Linguistic theories on foreign accent syndrome

The early explanatory reports on neurogenic FAS were mainly linguistically based. One of the first accounts explaining FAS after Whitaker formulated the diagnostic criteria, came from Graff-Radford et al. (1986). According to Graff-Radford et al. (1986) the FAS in their patient mainly affected the articulation of vowels due to increased tenseness in pharyngeal musculature and the vocal cords. However, they admitted that the **'tense posture hypothesis',** or more generally the '**phonetic setting hypothesis**' (see also Moen, 2000), was in their case purely based on the perceptual impressions of speech. No formal investigation of muscular tenseness were executed. The authors cannot explain why the patient had a tense speech musculature: the patient did not, for instance, demonstrate any upper motor neuron signs. The **advanced tongue root (ATR)** was forwarded as an alternative hypothesis. However, according to Ingram (1992) an advanced tongue root is associated with tense speech musculature (see also Ladefoged, 1971) and hence the issue remained unresolved.

Around the same time Blumstein et al. (1987) forwarded their **'timing control hypothesis'** or **'prosodic hypothesis'** (cfr. Monrad-Krohn, 1947). According to Blumstein et al. (1987) the segmental changes in FAS occur secondary to suprasegmental changes (e.g. vowel elongation can be explained by amongst other variables, a slow speech rate). In their opinion, FAS is primarily a disorder of melody and rhythm of language. Although she suffered myasthenia gravis the authors state that their patient was not dysarthric. Myasthenia gravis is associated with flaccid dysarthria and the muscle atrophy implies difficulty in reaching articulatory targets. However, the acoustic analysis of the patient's vowels did not reveal any major deviations compared native speakers of English. Still, the patient's accent was perceived by some as Slavic, or Romance. Because of the discrepancy between the perceptual impressions and the acoustic findings, Blumstein et al. (1987) argue that FAS must represent a '**generic foreign accent**' in speech that is not perceived as disordered, as would be the case for aphasia, apraxia of speech, or dysarthria.

Ingram et al. (1992) refined the hypothesis formulated by Graff-Radford et al. (1986) and argued that the raised muscular tension and associated excursions of articulators from neutral position when articulating segments can explain part of the segmental and suprasegmental changes. However, Ingram et al.'s (1992) patient demonstrated consonantal fortition in combination with weakening processes. Hence, the tense vocal tract setting seemed insufficient as an explanation to account for these opposing characteristics. They argued that the hypotheses by Graff-Radford et al. (1986) and Blumstein et al. (1987) are not mutually exclusive: tense vocal tract setting can affect timing and rhythm in speech as well.

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Sometime later, Kurowski et al. (1996) confirmed the assumptions of Ingram et al. (1992) and argued that FAS was not just due to tense vocal setting, nor to a primarily prosodic disorder (Blumstein et al, 1987): their patient did not demonstrate any major prosodic deficits, although he did show a strong tendency to centralize vowels. The stress and rhythm changes in their patient were too subtle to induce the phonetic changes at the segmental level. In contrast to what had previously been argued, they believed that the speech changes of their patient were the result of **aberrant shaping and setting of the tongue**, (esp. affecting the cardinal vowels) and can be linked to a tenser vocal tract setting (cfr. if tongue root is advanced: ATR - hypothesis; Graff Radford et al., 1986). Still, it remains problematic that contrasting characteristics seem to induce the same perceptual effect: whether they demonstrate tension (Graff-Radford et al., 1986) or laxation (Blumstein et al., 1987)[1] of the vocal tract, patients are perceived as foreign speakers. Because of the absence of a fixed constellation of symptoms that describes this group of patients, Kurowski et al. (1996) argue that FAS is not a syndrome; nor do the patients acquire an authentic foreign accent. They argue that FAS is characterized by a generic accent, following Blumstein et al. (1987). Some features are salient to one listener, but not to another. This explains the great diversity in the recognized 'foreign accents'.

Ten years later, in 2006, Mariën et al. were the first to present a detailed account on the hypothesized **role of the cerebellum in FAS**. Based on functional neuroimaging data, they argue that FAS is the direct result of a disruption of cerebro-cerebellar connections between the frontal brain areas (motor planning, programming and articulation) and the cerebellum. In their patient, they could directly relate the remission of FAS symptoms to a stabilization of perfusion in the cerebellum. Of course, the cerebellum fulfills an important role in rhythm and timing (cfr. timing hypothesis above). The potential cerebellar involvement also led researchers to draw back on the comparison Whitaker (1982) already established: the one between FAS and ataxic dysarthria. These disorders share an important amount of characteristics. Can this be due to a shared pathophysiological mechanism? Many studies follow, arguing for a central role of the cerebellum in FAS, although it remained unclear what the exact contribution of this structure to the disorder comprises (Miller et al., 2006; Mariën et al., 2007; Cohen et al., 2009; Dankovičová et al., 2011; Verhoeven et al., 2013).

In the same year, Moen (2006) (again) explained FAS in the context of **gestural phonology**. She already pointed out the usefulness of a non-linear phonological approach to explain FAS in her 2000 review (Moen, 2000). The gestures necessary to reach articulatory goals/targets are defined along "a subset of related tract variables" (Browman & Goldstein, 1992 in Moen 2006; p. 412). The gestures are specified into gestural scores. Based on the FAS patient's data she concluded that FAS was due to **abnormal scaling and incorrect phasing of glottal gestures to oral gestures**, explaining the in-

[1] In Graff Radford et al. (1986) and Blumstein et al. (1987) both patients were speakers of American English who were perceived as Scandinavian.

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correct voicing and devoicing of consonants, vowel shortening and lengthening, and the misalignment of tonal contours.

Lastly, in 2013, van der Scheer et al. forward the **fortition hypothesis** as an explanation for the perceived segmental and suprasegmental changes in FAS (see also Gilbers et al., 2013; Jonkers et al., 2016). They argue that FAS is the result of a lack of co-articulation and increased tenseness, which can explain for instance the relatively frequent perception of FAS speakers of Germanic languages as speakers of Romance languages. Latter languages (including for instance French, Italian and Spanish) are marked by a syllable-timed instead of stress-timed rhythm as well as less vowel reduction (Jonkers et al., 2016). These are characteristics that are frequently associated with FAS speech. However, the authors also argue that the perceived accent will be different as the acoustic changes induced by this increased force of articulation are salient to some listeners, though less so to others. They agree with Ardila et al. (1988) and Kurowski et al. (1996) that FAS is ultimately in the "'ears' of the beholder" (Kurowski et al., 1996, p. 24).

Important to note is that apart from the evident connection with the tense posture hypothesis, this theory can also be linked to the suggestions of Verhoeven & Mariën (2007) who argued that there is a tendency in FAS speakers to move towards phonemes that deliver increased (proprioceptive) feedback, which are, for instance, plosives, and will inevitably lead to some of the other phonetic characteristics described in the context of fortition (e.g. less co-articulation) (see 1.4.3).

1.4.2. Psycholinguistic/cognitive accounts on foreign accent syndrome

In 1998, Whiteside & Varley were the first to offer a **psycholinguistic (cognitive) account of FAS**. In their paper 'A reconceptualization of apraxia of speech' they argue that FAS may be a subtype of apraxia of speech. According to the authors, apraxia of speech is the result of disrupted access to, or storage of, motor plans for articulation. They question whether the characteristics displayed by the patients are the direct result of this disruption, or the rather the expression of real-time compensation. They relate the issue to a difference in indirect and direct encoding processes, based on the speech model by Levelt (1989; Levelt, 1992). In the case of direct phonetic encoding, there is direct access to the phonetic storage with associated codes for the corresponding movement plans. Every time the speaker employs codes from this storage he/she refines them via the feedback mechanisms integrated in the speech network and becomes more skilled. Consequently, use of these 'trained' forms will demand less cognitive load over time than a system requiring constant real-time encoding of phonetic sequences and the associated articulatory gestures. Naturally, the direct route is then used for high-frequency words and syllables whereas for low frequency words and syllables online encoding is required. Whiteside & Varley (1998) argue that apraxia of speech and FAS should be seen as the extremities of a continuum expressing the severity of motor planning disorders. In case of apraxia of speech, patients rely more heavily on the indi-

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rect encoding route because of this disruption in access to the stored verbo-motor patterns or possibly because brain damage interferes with the patients capacity to execute the real-time coding efficiently (which may be further complicated by disrupted feedback systems). In the case of FAS speakers, the patients seem to be more efficient in online encoding when needed, and would still have access to the direct encoding route. Compensation is better, although still not perfect.

1.4.3. The computational theories of foreign accent syndrome

In 2007, Verhoeven & Mariën give an incentive for a new computational account of the mechanisms responsible for the segmental and suprasegmental changes in FAS. In their detailed account of a 53-year-old Dutch patient with FAS (French accent) and a comorbid apraxia of speech, the segments of speech were much more affected than prosody. In terms of suprasegmental characteristics, only speaking and articulation rate were markedly different from normal rates for standard Dutch, whereas intonation was in line with expectations. Interestingly, Verhoeven & Mariën (2007) argue that most of the speech characteristics could be directly associated with the comorbid apraxia of speech:

"*the slower speech rate, more tense laryngeal setting and articulatory simplification (explaining e.g. the devoicing of fricative and occlusives and overshoot of fricatives, less coarticulation) could be explained as the patient seeking to realize phonemes that offer increased proprioceptive feedback*." (Verhoeven & Mariën, 2007; p. 222, our translation)

Feedback and feedforward mechanisms have been explained in several models. However, the Directions In Velocity of Articulators or the 'DIVA'-neural network model (Guenther, 1994; Guenther et al., 2006; Tourville & Guenther, 2011) is among the most widely employed and particularly suited to explain deficits in apraxia of speech because it skips the conceptualization and formulation steps that are seen in other models (e.g. Levelt, 1989), and focuses on the level of articulation.

The model describes speech production by relying on two subsystems: a feedforward control system and a feedback control system. Applied to speech development, the model can be explained as follows: the phonemes that were acquired at an early age are stored in speech sound maps (in the inferior frontal gyrus and ventral premotor cortex); they lead to the development of the feedforward control loop. Via the feedforward control, one is aware of the phonological results of a set of motor programs (the predictive component) that are transferred to the articulator position and velocity maps. When the initiation maps in the (bilateral) supplementary motor area (SMA) become active, these commands are released. In these commands the position of the different articulatory variables (jaw height, tongue height, tongue shape etc.) are encoded, which are translated into movements of the vocal tract (or "articulatory synthesizer", Guenther et al., 2006, p. 281). The speech sound maps define the auditory and

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somatosensory target maps in the feedback control system. When speech is articulated, the somatosensory and auditory-perceptual awareness, mediated by subcortical nuclei, activates the feedback control loop that allows for an update of the feedforward loop that will try to correct articulatory encoding to achieve these targets correctly next time. The inconsistency between trials in apraxia of speech reveals that correction for prediction fails. Tomasino et al. (2013) argue that FAS too can be interpreted as a disorder of, most likely, feedforward commands, although this can depend on the lesion location: disrupted feedforward control in case of cortical damage and disrupted feedback control in case of subcortical damage. For their patient, who had a tumor affecting the left prefrontal gyrus, where articulator and velocity maps are stored, it was argued that mainly feedforward control was disrupted.

Interestingly, the cerebellum plays an important role in "the learning and maintenance of feedforward motor commands" (Tourville & Guenther, 2011, p. 11). As it stands in close connection to the motor areas, and on the other hand receives input from the somatosensory cortex and auditory areas, it is attributed a cardinal role in feedforward motor control via supposed feedback motor learning in response to a miscalibration of intended movement or execution targets (Schmamann & Pandya, 1997). If this model is apt to describe FAS, it could possibly equally explain why Mariën et al. (2006) found a direct relation between cerebellar reperfusion and resolution of FAS in their patient.

1.5. OUTLINE OF THIS DISSERTATION

As can be derived from the subsections above, previous FAS research has especially focused on providing explanations for descriptions of acquired neurogenic FAS. In current dissertation we aim at giving a comprehensive neurolinguistic account of foreign accent syndrome, including the developmental and psychogenic subtype. To this purpose we will aim at resolving the following issues in the subsequent chapters:

1) Provide a comprehensive neurolinguistic description of neurogenic, developmental and psychogenic FAS as distinguished by Verhoeven & Mariën (2010a), based on a systematic research synthesis of previously published cases, as well as new case descriptions (Chapters 2-8).

2) Investigate at which level FAS causes disruption in the speech production process: is it a programming disorder or an execution disorder? Based on its established semiological resemblance with apraxia of speech and ataxic dysarthria, it is evaluated whether it should be considered a subtype of another motor speech disorder or should be conferred an independent status (Chapters 2-4)

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3) Investigate the role of the cerebellum in the pathophysiological explanation for FAS (cfr. Cole 1971; Whitaker 1982) (Chapter 2-4)

The current dissertation is divided into three parts: part I discusses neurogenic FAS, part II developmental FAS and part III psychogenic FAS. Each part consists of a series of chapters that are introduced below.

PART I - NEUROGENIC FAS

Chapter 2: **Neurogenic FAS – A review**

In the second chapter, the concept of 'neurogenic FAS' is analyzed in detail. We present a review of all published neurogenic cases dating from 1907 (first anecdotal report of a FAS case by Pierre Marie), until October 2016. The acquired, vascular FAS cases constitute the focus point of this chapter (mainly for the reason that these allow for the best lesion-behavior correlations). We analyze the reported lesion locations in relation to the segmental and suprasegmental deficits, the associated comorbid disorders, cognitive deficits as well as remission data. Prior analyses constituted an important landmark in providing a complete description of vascular FAS as well as arguing that FAS is dual disorder: one of planning and execution, possibly explaining the shared characteristics with apraxia of speech and (ataxic) dysarthria.

Chapter 3 **– The Posterior Fossa and Foreign Accent Syndrome: Report of Two New Cases and Review of the Literature**

On rare occasions, FAS has been reported after posterior fossa lesions. Usually only mutism and dysarthria are reported after posterior fossa damage. The posterior cranial fossa is the cranial cavity at the rear end of the skull and contains the cerebellum and the brainstem. This chapter includes a review focused on patients who developed FAS after posterior fossa damage (Cole et al., 1971, case 1 & 2; Dankovičová & Hunt, 2011; Tran & Mills, 2013; Cohen et al., 2008; Mariën et al., 2013), as well as a discussion of cases for which it is clear that posterior fossa structures played a cardinal role in entertaining or resolving the disorder (Hwang et al., 2001; Verhoeven & Mariën, 2002; Mariën & Verhoeven, 2007, case 2). Two new case studies are discussed in this thesis. The first one is a 44-year-old Flemish Dutch speaking monolingual man, who started speaking with a Hollandic-Dutch accent after incurring an ischemic stroke in the right pons. FAS was diagnosed in combination with a brainstem cognitive affective syndrome and apraxic agraphia. Symptom constellation was supported by the retained physiological deficits (SPECT scan). The second patient was a 72-year-old polyglot, British male with English as his first language. He suffered an infarction in the vascular territory of left posterior inferior cerebellar artery (PICA), and presented with isolated linguistic deficits (differential polyglot aphasia and FAS), severely affecting his Dutch, though not his first language. The diagnosis of FAS in the context of a left cerebellar damage (case 2) and

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hypoperfusion (case 1) is discussed in the context of a disruption of the delicate balance in the bi-hemispheric (cerebro-cerebellar) speech network.

PART II - DEVELOPMENTAL FAS

Chapter 4 – Developmental Foreign Accent Syndrome: Report of a New Case

In this chapter, a new case of developmental FAS is reported. The patient is a 17-year-old monolingual, Dutch-speaking Belgian boy, who has been speaking with a French accent as of language development. Clinical neurological evaluation and structural imaging via MRI did not demonstrate any abnormalities. Tc-99m-ECD SPECT disclosed a hypoperfusion at the level of the prefrontal and medial frontal regions, as well as in the lateral temporal regions and the right cerebellum. The neurolinguistic evaluation of the patient's speech and language revealed the existence of a comorbid developmental apraxia of speech. The patient demonstrated specific executive dysfunctions and a visuo-constructional dyspraxia in the presence of above-average intellectual and mnestic capacities. In the discussion the relation between the co-existing deficits is evaluated in the light of a disruption of the cerebro-cerebellar network subserving these functions.

PART III – PSYCHOGENIC FAS

Chapter 5 - Foreign Accent Syndrome As a Psychogenic Disorder: A Review

The aim of this first chapter of the third part of current thesis is to review the psychogenic FAS cases published until July 2014. Although it may be clear that the majority of the FAS cases acquired their 'foreign' accent after neurological damage, there has been incremental reporting of FAS cases for which a neurological explanation was either highly improbable or completely discarded. In this chapter, the instances of psychogenic FAS are analyzed and discussed in detail. Ample attention is devoted to demographic characteristics, associated psychological disorders, segmental and suprasegmental characteristics, onset and remission of the accent. The review provides a clear overview of the characteristics of reported psychogenic FAS patients in order to aid the diagnostic process and argues that the FAS criteria as formulated by Whitaker (1982) are insufficiently inclusive to capture all instances of FAS and hence are in need of revision.

Chapter 6 – Perceptual Accent Rating and Attribution in Psychogenic FAS: Some Further Evidence Challenging Whitaker's Operational Definition

In chapter 6, a new case of psychogenic subtype of FAS is presented. The patient is a 40-year-old, non-aphasic, polyglot (L1: French, L2: Dutch, and L3: English) woman from Belgium, with a 12-year history of addiction to opiates and psychoactive substan-

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ces, and psychiatric problems (including suspected conversion disorder). She presented with a foreign accent of sudden onset that only affected her L1, whilst other languages remained unaffected. There were other indications that the accent was developed on psychiatric grounds: during a temper tantrum in the hospital, the patient suddenly lost the accent. In addition to the outline of the diagnostic work-up in which we try to underline the characteristics that argue in favor of the psychogenic origin, the presence of the foreign accent was formally confirmed after a perceptual accent rating and accent attribution experiment.

Chapter 7 **– Psychogenic Foreign Accent Syndrome: A New Case**

In this chapter we report a 33-year-old, right-handed, French-speaking Belgian woman who was involved in a car accident as a pedestrian. Six months after the incident, she suddenly developed a German/Flemish-like accent. The psychological assessments as well as the clinical interview confirmed the presence of psychological problems, while neurological damage was excluded by means of repeated neuroimaging and neurological examinations. Interestingly, the accent fluctuated along with the patient's mood, and the accent onset was argued to coincide with the period when the patient was dismissed from work due to 'personality change' (she became blunt with customers, coworkers and her boss). As was the case for the patient described in chapter 6, a perceptual accent rating and attribution experiment was performed which disclosed that the patient was perceived as speaking with a foreign accent.

Chapter 8 **- Neologistic Jargon in a case of Psychogenic Foreign Accent Syndrome**

In chapter 8, we describe the case of a 28-year-old, monolingual, native Dutch-speaking woman from the Netherlands who developed a foreign accent (perceived as Flemish or German) two weeks after a fall down the staircase during her first pregnancy. Acute neurological examinations were normal and neuroimaging ruled out structural brain damage. The patient developed a FAS and displayed language mixing and language switching behavior whenever stress increased: Dutch was mixed with German(-like) words and/or sentences. Symptoms equally fluctuated with stress, fatigue or emotional load. When stressors were increased, she developed a transient neologistic jargon speech, a condition never observed before in the context of FAS. Psychodiagnostic tests and a clinical psychological interview revealed that there were no indications for psychopathy, although the accent was clearly induced when the examiner touched upon traumatic incidents or increased emotional stress. Moreover, there were reports of psychotic episodes in the medical history, and highly incongruent findings in the speech profile. In the discussion the diagnostic process of psychogenic disorders is critically evaluated, as it is – and always has been – the object of scientific scrutiny: it is largely based on clinical experience and exclusion criteria.

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Chapter 9 - GENERAL DISCUSSION

In chapter 9 we describe the most important findings from chapters 2-8 in conjunction with the theories and research aims formulated in chapter 1. This chapter provides a discussion of what characterizes neurogenic, developmental and psychogenic FAS in terms of demographics, etiology, associated neuropsychological issues, comorbid speech and language disorders, remission as well as segmental and suprasegmental characteristics. Based on the information in chapters 1-4 it will be argued that neurogenic FAS cannot be seen as a unitary concept at the level of planning of execution, but that there are arguments that favor a dual perception: FAS as at once a planning and execution disorder. This will be discussed in the context of the associated perceptual characteristics, lesion locations as well as comorbid cognitive disorders. Lastly, it will be discussed how the cerebellum may subserve a fundamental role in FAS. Not only when directly damaged (chapter 3), but also, and foremost, due to its connections with frontal areas (BA 4 and 6), which are usually the damaged areas in FAS.

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PART I: NEUROGENIC FAS

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CHAPTER 2

Foreign Accent Syndrome revisited: more than 100 years of mistaken identity

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ABSTRACT

FAS is a puzzling motor speech disorder that has attracted the attention of the scientific community and the media for more than a century. Currently no model exists that can single-handedly explain the pathophysiology of the disorder. In this article, a corpus of 172 FAS cases published between 1907 and October 2016 was collected and analyzed with respect to demographic and linguistic characteristics, associated neurocognitive symptoms, and comorbid disorders.

In line with psychogenic FAS, analysis of the study corpus disclosed that FAS more frequently affects women due to a neurological disorder (neurogenic FAS) than men. In neurogenic FAS, patients usually acquire FAS after a stroke, often in association with aphasia, dysarthria or apraxia of speech. FAS lesions are typically situated in the left supratentorial brain regions, generally involving the primary motor cortex (BA 4) and the basal ganglia. Neurocognitive symptoms have not been systematically looked for in FAS but when present, frontal executive functions are usually affected. In terms of FAS characteristics, the current review identified a diverse set of segmental and suprasegmental changes. At the segmental level, vascular FAS is primarily associated with consonant substitutions and vowel elongations. At the suprasegmental level, FAS principally induces a slow speech rate. A comparison with apraxia of speech (AoS) and ataxic dysarthria reveals that FAS resembles ataxic dysarthria on a suprasegmental level, whereas on the segmental level, the changes correspond to the (perceptual) diagnostic markers of AoS. On the basis of a close comparison of the cognitive and theoretical accounts of FAS, AoS and ataxic dysarthria, FAS can be viewed as a dual component motor speech disorder in which both planning and motor execution of speech are mildly affected. Analysis of the data further disclosed a fourth FAS variant that might be called the 'mechanic-organic variant'. In the absence of CNS damage, the mechanic-organic variant has been described after temporo-mandibular joint surgery, influencing the mechanics of the oral cavity. Finally, our review shows that Whitaker's (1982) widely used criteria fall short in encompassing all FAS patients. A need for a reappraisal is forwarded in which we suggest to broaden the definition of FAS to a neurological, psychiatric, mixed or mechanically induced motor speech planning and execution disorder in which segmental and/or suprasegmental changes lead to the impression of an altered accent within a group of interlocutors of the same linguistic community.

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2.1. INTRODUCTION

Foreign Accent Syndrome (FAS) is a relatively rare motor speech disorder characterized by speech errors that are perceived as a foreign accent by members of the same language community as the patient. As such, it is the only speech production disorder that is defined in perceptual terms, i.e. it explicitly refers to the auditory impression it creates in listeners.

A change of regional accent due to acute brain damage was described for the first time more than 100 years ago by the French neurologist Pierre Marie (1907), who described a patient whose original Parisian French accent had changed to what was perceived as Alsatian French after recovery from anarthria. Since Marie's anecdotal description of FAS, the condition has been documented in at least 172 case reports.

Notwithstanding ample attention of the scientific community, little progress has been made towards a fundamental understanding of this disorder. A hundred years of multidisciplinary research has neither been able to identify the pathophysiological substrate of this syndrome nor identify a coherent system in the speech errors that might separate FAS unambiguously from other speech and language disorders such as apraxia of speech (AoS), ataxic dysarthria, or even polyglot aphasia. The purpose of this review article is to bring together the available data about FAS and critically review the phenomenon by means of an analysis of cases reported since 1907.

2.1.1. Genesis of the concept FAS and the introduction of diagnostic criteria

On July 20, 1907 Pierre Marie presented in a short communication to the Société des Hôpitaux in Paris, four patients who had recovered from anarthria after a left hemisphere stroke involving the lenticular region. The second patient he discussed was a right hemiplegic Parisian man [1][2] who had not been able to speak for nine years after a subcortical left hemisphere stroke. When the anarthria receded he was not aphasic and could, as Marie (1907) stated, express his thoughts in sufficient detail. He also knew to read and write (with his left hand). However, as a sequela, the patient's speech contained a kind of Alsatian accent he did not present before the insult:

'A côté de cet Anarthrique typique, vous en voyez un autre qui, hémiplégique droit lui aussi, est resté neuf ans sans pouvoir parler, et qui, actuellement, a recouvré le langage et peut exprimer ses pensées avec tout le détail nécessaire; il lit et écrit fort bien (de la main gauche), chez lui le langage intérieur n'est pas sensiblement atteint. Mais de son anarthrie antérieure, il reste cependant des vestiges très appréciables consistant dans une sorte d'accent alsacien assez marqué qu'il n'avait pas autrefois, puisqu'il est Parisien, et qui ne lui est venu que depuis qu'il a recommencé à parler.' (p. 109)

[2] Case descriptions in square brackets can be consulted in the Appendix A1

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The remarkable phenomenon of a change of regional accent during recovery from subcortical left hemisphere stroke was not discussed and Marie did not explicitly identify any of the speech characteristics with which this perceived change in accent may have been associated. By contrast, the association between the perceived accent and the patient's pronunciation characteristics was explicitly addressed in 1919 by the Czech neurologist Arnold Pick who published a case study involving a 26-year-old Czech butcher [2] who had developed a Polish accent after a left-hemisphere stroke. Pick identified the determining speech characteristics to be both segmental (the softer articulation of the Czech fricatives) and suprasegmental (wrong placement of word stress). However, in contrast to Marie's (1907) report mentioning 'a kind of' regional accent, Pick's (1919) extensive description may be considered a less pure and even ambiguous account of FAS after stroke. Indeed, the patient was stationed for several months in Poland as a soldier during the First World War and also presented aphasic symptoms such as agrammatism, paraphasias, perseverations, alexia and agraphia. As such, the change to a genuine Polish accent might also be regarded a neurologically induced shift to a previously learned foreign accent (Polish) as part of bilingual or polyglot aphasia reflecting a selective disruption (at the phonetic level) of the cognitive-linguistic system to control the correct selection of suprasegmental speech features of a particular language (accent).

A next landmark case description was that of the Norwegian neurologist Monrad-Krohn (1947) who reported the case history of the 30-year-old Astrid L [3]. She developed a German accent after traumatic head injury to the left fronto-temporo-parietal region following a fall down a steep incline when hit by a bombshell. Many Norwegians identified her accent as German and, in post-war Norway this acutely exposed the impact of such a change of accent on the patient's identity in society. Due to the German occupation of Norway during wartime, the patient "complained bitterly of constantly being taken for a German in the shops, where consequently the assistants would sell her nothing" (p. 410). According to Monrad-Krohn, this change of accent was predominantly related to suprasegmental problems pertaining to the disappearance of the Norwegian tonal distinction. This led to the longstanding view that FAS is essentially an 'ataxia of the prosodic faculty'.

Contrary to Monrad-Krohn's (1947) view on FAS as a prosodic disorder, Nielsen & McKeown (1961) regarded the condition as a peculiar form of dysarthria characterized by a disturbance of rhythm. They explicitly drew attention to the highly subjective interpretation of the 'dysprosodic qualities' of the syndrome [4-5]:

'(...) the dysprosody is simply interpreted as dysarthria unless it reminds the listener of some particular foreign language, and that therefore the interpretation is largely subjective. If the examiner did not have acquaintance with any language but his own, he would not see a simulation of some foreign language but only a peculiar form of dysarthria, a disturbance of rhythm' (p. 159).

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Critchley (1962) for the first time distinguished two categorically different types of accents: 1) intensification of a regional accent in aphasia due to removal of inhibitory factors as the result of disease and 2) appearance of something closely akin to a regional accent after a 'cerebro-vascular catastrophe' (post-aphasic dysprosodia). As an exception to the findings in the early contributions that explicitly linked FAS to vascular or traumatic damage of the language dominant left hemisphere, Critchley (1962) reported two cases with an atypical etiology and unique anatomoclinical profile. He described a 49-year-old woman (Case 1) [6] who in the absence of any clinical evidence for a structural brain lesion developed a regional accent in the context of a principal post-traumatic neurotic syndrome. The Welsh accent she presented aroused considerable interest in the media when she claimed damages in Court on account of 'the handicap' of a Welsh accent. Aphasic mutism followed by a regional Welsh accent in Critchley's third case did not result from left hemisphere damage but was due to a right hemisphere stroke. As this 37-year-old woman was said to be right-handed, this case (Case 3) [8] might be considered the first description of 'crossed FAS' due to reversed cerebral language dominance as reflected by the accompanying (crossed) aphasia.

Cole (1971) [13-14] further expanded the diversity of possible lesion locations in FAS beyond the supratentorial brain regions by presenting two patients in whom the causative lesion was situated in the posterior fossa. Former editor-in-chief of Neurology, Robert Joynt, ironically commented on this unexpected finding, suggesting a crucial role of the cerebellum in the integration of motor speech characteristics and the pathophysiology of FAS:

'...it doesn't appear unusual that any of the brain mechanisms which are used to integrate motor acts may alter force, prosody, and rhythm of speech. It is well that Dr. Cole has pointed this out with posterior fossa lesions. However, I am enough of a romanticist to think that these subtle inflections and nuances of speech must ultimately stem from the cerebral cortex. For example, if Juliet had said in a monotone, ''Romeo, Romeo, wherefore art thou, Romeo?'' I doubt if Romeo would have done himself in for her. But then, I don't think Shakespeare wrote for the cerebellum.' (p. 153).

In 1982, Whitaker coined the term 'Foreign Accent Syndrome' to denote the phenomenon in which a cerebral lesion induces a change of accent and for the first time defined the condition on the basis of a set of four operational criteria: 1) the accent is considered by the patient, by acquaintances and by the investigator to sound foreign, 2) it is unlike the patient's native dialect before the cerebral insult, 3) it is clearly related to central nervous damage (as opposed to an hysteric reaction, if such exists), and 4) there is no evidence in the patient's background of being a speaker of a foreign language (i.e., this is not like cases of polyglot aphasia).

To facilitate the classification of FAS cases, Verhoeven & Mariën (2010a) made a fundamental taxonomic distinction between neurogenic, psychogenic and mixed type FAS. In neurogenic FAS the development of the accent is related to organic damage to the

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central nervous system (acquired, congenital or developmental), while this is not the case in psychogenic FAS: in the latter the accent is grounded in psychogenic disorders. When the speaker with neurogenic FAS manipulates the accent to ensure greater accent consistency to create a more authentic foreign personality the term mixed FAS is applied.

2.2. METHODS

The relevant literature on FAS was identified through searches of scientific electronic databases (*Medline, PsycINFO, Current Contents, Web of Science*). In addition, bibliographies of all relevant articles were scanned to identify additional references. Only first source information was analyzed, second- or third-line references to original contributions were not taken into consideration. This resulted in a database consisting of 172 cases of FAS.

Quality of the collected data was variable in terms of detail. In order to make sound analyses, inclusion and exclusion criteria were applied to narrow down the corpus for further analysis. In terms of article type, the authors decided that cases reported in poster or oral presentations, and conference proceedings (e.g. 29, 30, 32, 33, 37, 42, 50, 51, 52, 79, 86, 107, 119, 120, 162) as well as unpublished theses (100-103) were to be excluded from further analysis. For the sake of completeness, the collected data can be consulted in the appendix (**Appendix A1**).

Inclusion criteria were broadly defined as (i) the description of a patient in whom a change of accent was established, (ii) and for whom the authors had a clear-cut standpoint with respect to the associated etiology in order to limit suggestiveness and misinterpretation of the data at hand.

This led to the supplementary exclusion of case 36, 39, 40, 68, 71, 81, 88, 126, 137, 158-161, 172. These cases were excluded for the following reasons: (i) formal statement that the etiology was unclear, or could not be identified (e.g. 36, 88), (ii) etiology was left out of the case description (158-161), (iii) hesitation with respect to a functional and/or neurological origin, leading to an absence of etiological classification ('suggestive') (39, 40, 81=121, 126, 137, 172). Due to linguistic constraints, only articles published in English, Spanish, Norwegian, German or Portuguese were included. One case had to be excluded because a translation could not be obtained: Tokudo et al. (2015, case 164).

Some of the cases in the corpus were reported more than once [10=6, 11=7, 12=8, 58=20,64=38,65=39,72=21,88=114,99=89;105=59,121=81;132=106=85=77=69=50=43, 133=78, 134=63, 135=94, 147=70=33, 151=115=100, 152=116=101, 153=117=109=102, 154=118=103]. When taking into account only 'authentic' cases that fulfilled the inclusion criteria, the number of included cases amounted to 112 cases [1-9, 13-28, 31, 34, 35, 38, 41, 43-49, 53-57, 59-63, 66, 67, 70, 73-76, 78, 80, 82, 83, 87, 89-98, 104, 108-113, 115, 116, 118, 122-125, 127-131, 136, 138-146, 148-150, 155-157, 163, 165-171] published since the first description in 1907 and October, 2016.

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2.3. RESULTS

2.3.1. Demographic characteristics

The demographic, etiological, neuroradiological, linguistic and neuropsychological characteristics recorded for the cases selected for the purpose of this study are listed in the table in the appendix (A1) and marked with an asterisk.

The large majority of the patients in the study corpus (97.32%) are adults (*n*=109/112); only three patients are children (<18 years) [51, 95, 168]. Seventy-five out of 112 patients are women (*n*=75/112; 66.96%), thirty-seven are men (*n*=37/112; 33.04%). A binomial test indicated that the proportion of female FAS patients was significantly higher than the proportion of male patients ($p<0.000$ - binomial test).

The mean age of the study population was based on calculations for 111 patients as age was not mentioned for the patient described by Marie (1907). The mean age of FAS patients was 47.93 years (range 7 to 88 years; SD: 14.59). Mean age of the female patient group (*n*=75) was 49.71 years (range 24 to 88 years, M=49; SD=12.73), mean age of the male patient group (*n*=36) was 44.22 years (range 7 to 76 years, M=46; SD=17.48). The difference between the mean age for men and women was not significant (*n*=111, *U*=1137.50, *p*=.181).

Hand preference was specified in 62/112 cases (55.36%). Fifty-nine out of the 62 cases were dextral patients (96.72%) [4, 5, 8, 16-20, 22-25, 28, 31, 38, 43, 44, 46, 48, 59, 61, 63, 66, 70, 74, 75, 76, 78, 80, 91, 92, 93-97, 99, 104, 109-112, 115, 116, 118, 123, 125, 127, 128, 136, 138, 140, 143, 144, 157, 166-170]. Three patients are left-handers [27, 47, 89]. For the remaining 50 patients, handedness was not specified. Right/left hand dominance ratio was approximately 21:1. Handedness was formally assessed by means of a questionnaire or handedness test in twelve of the right-handed cases [17, 43, 44, 63, 70, 78, 94-96, 138, 166, 167] (*n*=12/62, 19.35%) and in one of the left-handed patients [89].

Twelve out of the 112 cases (10.71%) were described as bilingual[3] [2, 7, 16, 27, 34, 67, 92, 98, 111, 125, 127, 136] and fourteen were polyglot patients [13, 14, 31, 43, 46, 60, 63, 80, 94, 112, 145, 166, 167, 169] (12.5%). Thirty patients were said to be monolinguals [4, 15, 18-20, 59, 70, 73, 78, 89, 91, 95, 104, 109, 115, 116, 118, 130, 131, 138-140, 143, 144, 146, 148-150, 163, 170] (*n*=30/112; 26.79%). For 109 patients (*n*=109/112; 97.32%) the native language was documented in the case report, but for 3 patients this was not done [35, 155, 156].

[3] The authors are aware that concepts such as 'bilingualism' and 'polyglot' can be defined in different ways. For the purpose of establishing this corpus all types (compact, coordinated, subordinated, balanced, dominant - Fabbro, 2003) were included, without making distinctions. We followed the authors' descriptions in detail.

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As can be seen in appendix A1, the large majority of the patients were native English speakers (*n*=60/112, 53.57%). In twenty-eight patients (25%) there was proof that the patients definitely had had prior contact with the accent that became their 'FAS-accent' [2, 5, 6, 7, 13, 14, 27, 34, 43, 53, 54, 60-63, 90, 98, 109, 111, 112, 125, 128, 129, 130, 136, 143, 169, 170]. In 46.43% (*n*=13/28) of these cases, there was a clear reversion to a previously learned accent, as these patients were either bilinguals or polyglots that spoke the language associated with their FAS-accent [7, 13, 14, 34, 43, 60, 63, 98, 111, 112, 125, 136, 169]. Others also had early contact with the accent due to language contact with relatives [2, 5, 6, 27, 53, 54, 61, 90, 109, 128, 129, 130, 143] or had had contact with the accent due to formal language instruction at school at a later age [62, 170]. Mind that for the developmental FAS cases discussed below, the patients learned French *after* they developed the French-like accent in the context of FAS [94, 166-168].

2.3.2. Etiology

76.79% of the authentic (de novo) case descriptions represent instances of acquired neurogenic FAS (*n*=86/112) [1-5, 7-9, 13-28, 31, 34, 35, 38, 43-49, 53-57, 59-61, 66, 67, 70, 74-76, 78, 80, 87, 89, 91-93, 96, 104, 108, 109, 111-113, 115, 116, 118, 122, 123, 125, 127, 128, 131, 136, 138-140, 143-146, 148-150, 155, 156, 163]. Five patients (4.46%, *n*=5/112) developed FAS as a developmental disorder [94, 95, 166-168]. Eighteen patients (16.07%; *n*=18/112) developed FAS in relation to a psychological or psychiatric disorder [6, 41, 62, 63, 82, 83, 90, 97, 98, 110, 124, 129, 130, 157, 165, 169-171], one patient suffered mixed FAS after stroke [73]. Two patients could not be categorized in the subtypes as described by Verhoeven & Mariën (2010a): case 141 and 142, described by Dilollo et al. (2014), who developed FAS on a non-neurological basis after a TMJ surgery. Their FAS could not be directly related to psychological disturbances. Rather, case 141 experienced some psychological and psychosocial changes *after* the onset of her FAS, and travelled to England where she felt she would be more at ease with the newly adopted accent. The authors do not infer whether FAS was completely responsible for these psychological changes, as is the case in mixed FAS.

Fifty-nine patients of the study corpus (*n*=59/112; 52.68%) developed vascular FAS due to stroke [1, 2, 4, 7, 8, 13-22, 24, 27, 28, 31, 34, 38, 43, 44, 48, 49, 54, 55, 59, 61, 66, 67, 70, 73-75, 78, 89, 93, 96, 108, 109, 111, 112, 115, 116, 118, 122, 123, 125, 127, 131, 136, 139, 140, 143, 145, 146, 149, 163] (**Figure 2.1**). Twenty-two of the fifty-nine stroke patients are men (37.29%) and thirty-seven are women (62.71%), although women are not significantly more affected by vascular FAS than men (*p*=0.067). Mean age of the total vascular group is 51.57 years, excluding the case of Pierre Marie [1], for which age was not provided (range= 24-76 years, M= 53, SD= 12.51 years). A trend was found for men (mean age=50.14 years, M=53 years, SD=14.40) to be younger than women (mean age=52.38 years, M=53 years, SD=11.43 years), but the difference was not statistically significant (t(58)=-,651; *p*=0.518).

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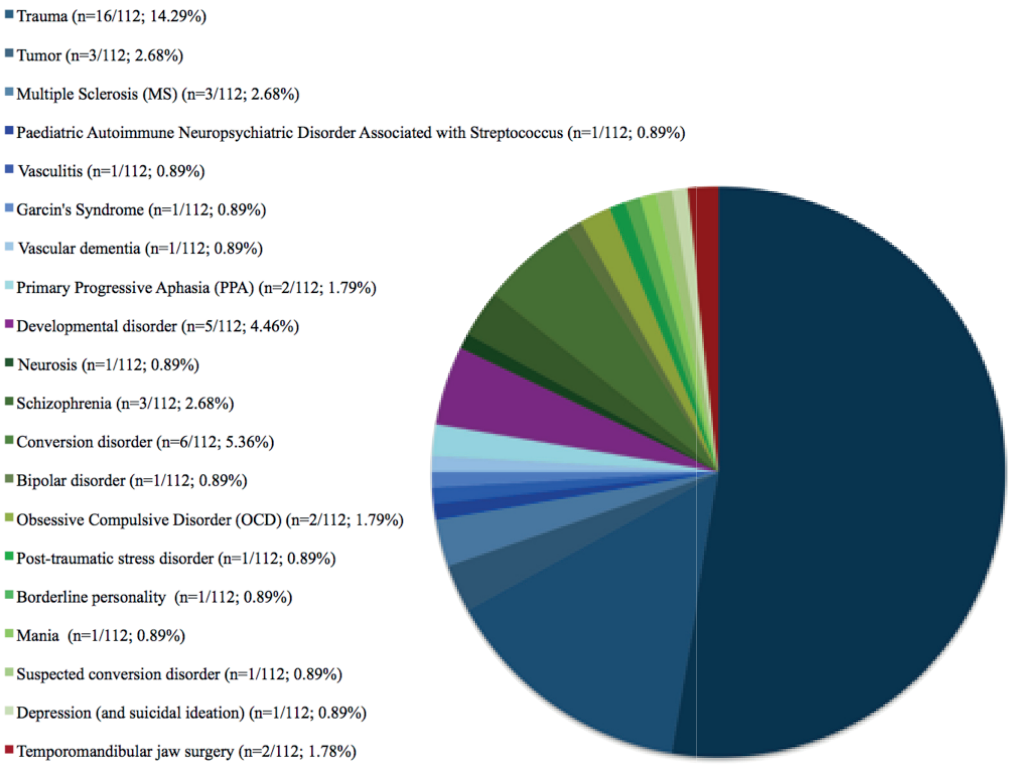
Figure 2.1. Pie chart with slices representing the different etiologies (in percentages). Mind the color coding: **neurogenic** FAS = in **BLUE** gradient, **developmental** FAS = in **PURPLE**, **psychogenic** FAS = in a **GREEN** gradient, **non-neurological** cases = in **RED**. The legend is included next to the pie chart.



In sixteen patients (*n*=16/112; 14.29%) FAS resulted from a cranio-cerebral trauma [3, 5, 9, 23, 25, 26, 35, 46, 53, 56, 57, 60, 104, 144, 155, 156]. Together with stroke-based FAS, they make up the gross of the associated etiologies. Four patients (*n*=4/112; 3.57%) presented with FAS in the context of an inflammatory disease such as multiple sclerosis (hereafter: MS) [45, 47, 92] or vasculitis [148]. In three patients (*n*=3/112; 2.68 %) FAS was identified during the course of a primary neurodegenerative disorder [76, 80, 128]. In three other cases (*n*=3/112; 2.68%), a brain tumor caused FAS [91, 113, 138].

In five cases (*n*=5/112; 4.46%) FAS occurred as a neurodevelopmental disorder [94, 95, 166-168]; sometimes co-occurring with another, associated neurodevelopmental speech/language disorder [94,168: developmental AoS; 95: Specific Language Impairment].

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In one patient [150] (*n*=1/112; 0.89%) the infection considered to have incited the FAS was identified as Paediatric Autoimmune Neuropsychiatric disease associated with Streptococcal infections (PANDA). The disease is known to mimic symptoms of (neuro/pedo-)psychiatric disorders such as Obsessive-Compulsive Disorder (OCD) and Attention and Hyperkinetic Disorder (ADHD). In one patient (*n*=1/112; 0.89%) a multifactorial cause (trauma, viral infection: Garcin's syndrome) was suspected [87]

Keeping in line with the inclusion and exclusion criteria, eighteen patients (*n*=18/112; 14.35%) could be classified as suffering from psychogenic FAS. Six presented with FAS in the context of a conversion disorder (*n*=6/112; 5.38%) [63, 90, 97, 98, 110, 157], three in the context of schizophrenia [41, 82, 165] (*n*=3/112; 2.68%), one in the context of a bipolar disorder (*n*=1/112; 0.89%) [83], one in the context of a neurosis (*n*=1/112; 0.89%) [6], one in the context of mania (*n*=1/112; 0.89%) [124], two in the context of obsessive-compulsive disorder (*n*=2/112; 1.87%) [129, 130], one in the context of post-traumatic stress disorder (*n*=1/112; 0.89%) [171] and finally, three in the context of an unspecified psychiatric disorder (*n*=3/112; 2.68%) [62, 169, 170]. Case 62 demonstrated depressive symptoms and suicidal ideation, case 169 fulfilled some of the criteria of conversion disorder but a formal diagnosis could not be made, and case 170 had a borderline personality, but a formal diagnosis of clinical disorder could not be made on DSM axis 1 and 2. However, the authors remain certain that these psychiatric issues played a cardinal role in the onset of FAS in these patients (for a more in-depth discussion of psychogenic FAS see Keulen et al., 2016d).

2.3.3. Lesion lateralization in FAS

Table 2.1. Structural brain damage in Foreign Accent Syndrome. The left hand column indicates the cases for which brain damage affected the left hemisphere, the right hemisphere, both hemispheres, cases for which there was no indication as to lateralization, or when the authors claimed there was no visible structural damage. The top-hand row indicates cases for which the lesion was supratentorial, infratentorial, both supra- and infratentorial, for which cases there was no indication as to position compared to tentorium, or the authors argued there was no perceivable structural damage. When neuroimaging was available, cases were marked by an asterisk. The cases can be verified in the table in the appendix to chapter 2 (A1).

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	Supra-tentorial	Infra-tentorial	Both infra-and supratentorial damage	Not indicated	No perceivable structural lesions	Total
Left	**61**	**2**		**3**		**66**
	1-5,7,9*,15-17*,19*,20*,21,22*,24*, 25*,26*,27*,28,31,34*,35*,43*,47*,48*,49*,53,54,55,57,59*,60*,61*, 66*,73*,74*,75*,76*, 78*,89*,91*,92*,93*, 96*,108*,111*,112*, 113*,118,122*,123*, 127*,128*,131*,136*,138*,140*,144*,155*,163*,166*	109*,139*		56,115,116		
Right	**3**	**1**		**1**		**5**
	23*,38*,70*	125		8		
Bilateral	**6**		**2**			**8**
	18,46*,80*,104*,143,156*		45*,65*			
Not indicated	**1**	**2**	**1**	**5**		**9**
	44*	13,14	145*	67,146,148,149, 150		
No perceivable structural damage					**24**	**24**
					6,41*,62*,63*,82*,83*,87*,90*,94*,95*,97*,98*,110*,124*,129*, 130,141*,142*,157*,165*, 168*,169*,170*,171*	
TOTAL	**71**	**5**	**3**	**9**	**24**	**112**

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As shown in **table 2.1** the majority of FAS patients had damage situated in the supratentorial left hemisphere (*n*=61/112; 54.46%) [1-5, 7, 9, 15-17, 19, 20-22, 24, 25-28, 31, 34, 35, 43, 47, 48, 49, 53, 54, 55, 57, 59, 60, 61, 66, 73, 74, 75, 76, 78, 89, 81, 92, 93, 96, 108, 111, 112, 113, 118, 122, 123, 127, 128, 131, 136, 138, 140, 144, 155, 163, 166]. In line with the previous publications on psychogenic FAS, no structural damage was reported in psychogenic FAS cases [6, 41, 62, 63, 82, 83, 90, 97, 98, 110, 124, 129, 130, 157, 165, 169, 170, 171]. For case 141 and 142 an MRI was inconclusive due to artifacts from metal implants in the jaw region.

68.85% (*n*=42/61) of the patients with lesions situated in the supratentorial left hemisphere, were stroke patients [1, 2, 4, 7, 15-17, 19, 20-22, 24, 27, 28, 31, 34, 43, 48, 49, 54, 55, 59, 61, 66, 73, 74, 75, 78, 89, 93, 96, 108, 111, 112, 118, 122, 123, 127, 131, 136, 140, 163]. Of note, 59.52% (*n*=25/42) of these stroke patients were right-handed, 4.76% (*n*=2/42) were left-handed and for 35.71% (*n*=15/42) handedness was not indicated. Right hemisphere damage occurred only in FAS cases who suffered from stroke [8, 38, 70, 125] (3.57%, *n*=4/112). Cases with damage restricted to infratentorial regions all suffered from stroke. All of the patients in latter categories were right-handed, and for case 139 handedness was not indicated. For cases with infra- and supratentorial damage, only case 45 had not suffered a stroke. The patient's T2-MRI showed hyperintensities at the infra- and supratentorial level and she was diagnosed with MS.

For many of the early studies the etiology was often based on neurological signs, and neuroimaging was not reported. For 75 % (*n*=84/112) of the cases information on brain imaging was provided (indicated with an asterisk in **table 2.1** above).

Until 1983 (Schiff et al.), neuroimaging was performed for only one patient in the corpus, described by Whitty (1964) [9], who reports the findings on a cerebral angiogram (technique first described by Egas Moniz, 1927) in a patient who developed FAS after the excision of an arteriovenous malformation. Between 1983 and 1992 (case Seliger et al., 1992; case 27) scans were exclusively done by means of computerized tomography (CT) [9, 16-20, 22-26]. Afterwards, CT was often substituted or complemented by an MRI [27, 28, 35, 41, 43-48, 59, 61, 63, 66, 67, 73-76, 82, 83, 87, 89-98, 108-111, 113, 127-129, 136, 138-141, 144, 155, 156, 157, 165-171] and/or DTI [92, 123, 127, 162, 163]. Functional neuroimaging (fMRI/SPECT) was only occasionally performed usually serving diagnosis with the patient in resting state [43, 76, 78, 90, 92, 94, 95, 113, 123, 127, 128, 138, 163, 168], but also for very specific scientific purposes, while the patient performed a task (picture naming) to disclose the changes in the speech network [59]. In three cases [91, 113, 138], the patient developed FAS due to a neoplasm and underwent direct electrical stimulation for identification of eloquent brain areas. This process is common for tumoral resection in language-related brain areas under awake surgery (De Witte & Mariën, 2013).

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2.3.4. Lesion location in vascular FAS

A number of studies have shown that the most appropriate etiology for establishing anatomoclinical correlations are circumscribed cerebrovascular lesions (including infarcts, hemorrhages and ruptured aneurysms with intraparenchymal damage) in an intermediate period ranging from about three weeks to about three months post-onset (Alexander, 1989; Mazzochi & Vignolo, 1979). The morphology of the lesion remaining the same, neurobehavioral symptoms in the earlier period (the *acute phase*) are often more severe than the lesion *per se* would entail [due to the additional effect of diaschisis affecting ipsi- or contralateral brain areas, mass effect and perilesional damage (penumbra)]. The effects of damage also depend on the affected brain area (Witte et al., 2000) and they may become less severe in the later period (the *chronic phase*) due to the opposite effect of functional compensation resulting from spontaneous recovery or induced by therapy. Neurocognitive and neurolinguistic disorders presumably mirror the effect of the lesion most faithfully in this intermediate period – called the *lesion phase* of the stroke by Alexander (1989) – when the lesion is stabilized, associated neurobiological phenomena disappear and compensation is minimal (Mazzochi & Vignolo, 1979). Appendix A1 shows that in vascular FAS patients, lesions are often situated in and around BA 4 and 6 (SMA, premotor area and motor strip) [16, 17, 22, 28, 43, 74, 93, 108, 122, 143, 163] and less often BA 44/45 (Broca's area) and frontal operculum [19, 43, 48, 63, 127] and much less often Wernicke's area [111].

To establish lesion behavior correlations for FAS in the present review, care has been taken to single out the stroke cases (*n*=59/112, 52.68%). These case reports were scanned in order to collect information regarding the time of onset of FAS. 54 of the 59 case reports with sufficient information to situate FAS onset in either the acute, lesion or chronic phase were included for further analysis (9.15%, *n*=54/59) [1, 2, 4, 8, 9, 14-22, 24, 27, 28, 31, 34, 38, 43, 44, 48, 49, 54, 55, 59, 61, 66, 67, 74, 75, 78, 89, 93, 96, 101, 108, 109, 111, 112, 116, 118, 122, 123, 125, 127, 131, 136, 139, 140, 143[4], 145, 163]. Results demonstrated that forty-one of the stroke patients (*n*=41/59; 69.49%) presented with FAS in the acute phase of the stroke [4, 9, 14, 15, 17, 18, 20, 21, 22, 27, 28, 43, 44, 48, 54, 55, 59, 61, 66, 67, 74, 75, 78, 89, 93, 96, 101, 108, 109, 111, 118, 122, 123, 125, 127, 136, 139, 140, 143[5], 145, 163], nine cases (15.25%) developed FAS in the lesion phase of the stroke [16, 19, 24, 34, 38, 49, 112, 116, 131] and four cases (6.78%) developed FAS in the late phase of the stroke [1, 2, 8, 31]. For the remaining five patients (8.47%) FAS onset could not be situated at all in relation to stroke onset [7, 13, 70, 146, 149].

[4] FAS based purely on self-report (Berthier et al., 2015). FAS was no longer present at the time the researchers saw the patient.

[5] FAS based purely on self-report (Berthier et al., 2015). FAS was no longer present at the time the researchers saw the patient.

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Eighteen of the 41 patients (*n*=18/41; 43.9%) with FAS in the acute phase still had accented speech in the lesion phase of the stroke [22, 27, 43, 44, 54, 55, 59, 61, 75, 78, 93, 109, 118, 122, 125, 136, 143, 163].

After verification of the imaging data, it was concluded that eight cases had damage largely restricted to the subcortical areas [27, 54, 55, 59, 75, 78, 109, 122]. Thirteen of the 18 patients were right-handed [22, 43, 44, 59, 61, 75, 78, 93, 109, 118, 125, 136, 143] (*n*=13/18, 72.22%), one patient was left-handed [27] (*n*=1/18; 5.56%) and for the remaining four patients handedness was not indicated (*n*=4/18; 22.22%). In fifteen patients in whom FAS persisted throughout the lesion phase, the condition resulted from a left hemisphere lesion [22, 27, 43, 54, 55, 59, 61, 75, 78, 93, 109, 118, 122, 136, 163] (*n*=15/18; 83.33%). The three remaining cases developed FAS after an infarction in the body of the corpus callosum [44], a hemorrhage in the right cerebellum [125], and bilateral hemorrhages affecting the left motor cortex, right insula and putamen [143].

2.3.5. Comorbid speech and language disorders in vascular FAS

In the stroke population [1, 2, 4, 7, 8, 13-22, 24, 27, 28, 31, 34, 38, 43, 44, 48, 49, 54, 55, 59, 61, 66, 67, 70, 73-75, 78, 89, 93, 96, 108, 109, 111, 112, 115, 116, 118, 122, 123, 125, 127, 131, 136, 139, 140, 143, 145, 146, 149, 163] (*n*=59), 40.68% (*n*=24/59) of the cases were mute in the acute phase, before they developed FAS [1, 2, 4, 8, 15, 16, 19, 20, 22, 24, 28, 34, 43, 61, 73, 74, 78, 96, 116, 118, 136, 140, 143, 145].

Aphasia was noted in the acute phase in nineteen stroke cases [2, 4, 15, 16, 17, 24, 31, 44, 48, 49, 61, 66, 67, 75, 78, 112, 122, 136, 163] (*n*=19/59; 32.20%). Two cases developed aphasia in the lesion phase [19, 34] (*n*=2/59; 3.39%), and one case in the late phase [131] (*n*=1/59; 1.69%) of the stroke.

Dysarthria was found in the acute phase in eleven stroke patients [14, 17, 20, 22, 27, 31, 49, 89, 96, 125, 136] (*n*=11/59; 18.64%), and in three patients dysarthric speech problems occurred in the lesion phase [16, 38, 131] (*n*=3/59; 5.08%). Case 13 also suffered comorbid dysarthria. However, the onset of the FAS and dysarthria could not be estimated as it was not clear when the cerebellar anoxia had occurred and whether the patient had immediately gone to the hospital.

Apraxia of speech was reported in the acute phase in six stroke patients (*n*=6/59; 10.17%) [15, 43, 75, 93, 131, 136] and all of these patients, except case 131, also suffered FAS in the acute phase. For one patient AoS was noted in the chronic phase (tested 26 months post-onset) [115]. However, it is not clear whether the patient presented AoS before that stage.

Agrammatism occurred in ten stroke patients [2, 15, 16, 18, 19, 55, 78, 112, 136, 143, 163] (*n*=11/59; 18.64%). Roth et al. (1997) [34] provide contradictory information concerning their patient, claiming once that their patient was agrammatic (p. 551) without providing evidence for the grammatical disorder, and stating that "the language of [their] patient was grammatically correct and lexically full" (p. 550). Due to the conflicting information, this case was not taken into consideration. In three patients

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[16, 19, 78] agrammatism developed in the lesion phase (*n*=3/59; 5.08%). Two stroke patients presented with agrammatism without aphasia [18, 55] in the acute stage (*n*=2/59; 3.39%), and one patient was described as producing 'grammatical errors', without any further indications [74] (*n*=1/59; 1.69%). This patient was not aphasic and did not suffer any other comorbid speech or language disorder, except for a pre-FAS mutism.

Alexia was attested in four patients in the acute phase [2, 8, 44, 96] (*n*=4/59, 6.78%). Agraphia was noted in the acute phase in five stroke patients [2, 4, 8, 44, 96] (*n*=5/59, 10.17%) and in the lesion phase in one patient [118] (*n*=1/59, 1.69%).

2.3.6. Speech characteristics of vascular FAS

Table 2.2. Characteristics of vowels in patients with vascular FAS

VOWELS		
Type	**Case**	**Total**
Raising	15,17,34,38,89,96,109,112,122,163	10
Lowering	19,21,22,89,127	5
Fronting	15,17,19,34,38,74,96,109	8
Backing	17,19,27,34,74,109,122,163	8
Elongation	15,17,18,27,35,38,48,74,75,89,109,118,123, 131,140	15
Epenthesis (schwa)	20,31,74,75,89,109,139	7
Shortening	2,21,74*,75*,109,163	6
Omission	19,22,78,109,163	5
Diphthongization	19,59, 89,131,163	5
Monophthongization	15,38,74,109	4
Addition	19,34,138	3
Delabialization of rounded vowels	21	1
Excessive rounding	38	1
Laxation	31	1
Centralization (compression of vowel space)	31	1
Offglides	59	1
Metathesis	22	1
'Substitutions' (unspecified)	75	1

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Vowel changes characterizing speech in vascular FAS patients are presented in **Table 2.2** above. FAS speech is foremost characterized by elongations (vowel lengthening) (*n*=13/59, 22.03%), a tendency towards F1 and F2 changes (raising, lowering, fronting and backing), and epenthesis. Raising [15, 17, 34, 38, 89, 96, 109, 112, 122, 163] and lowering [19, 21, 22, 89, 127] (F1 movement, *n*=14/59, 23.73%) as well as fronting [15, 17, 19, 34, 38, 74, 96, 109] and backing [17, 19, 27, 34, 74, 109, 122, 163] (F2 movement, *n*=11/59, 18.64%) occurred in the speech of respectively almost a fourth of the vascular FAS patients. Changes affecting raising and lowering (F1) (case 87) and especially fronting and backing (F2) (cases 17, 19, 34, 74, 109) sometimes co-occurred.

Table 2.3. Characteristics of consonants in patients with vascular FAS

CONSONANTS		
Type	**Case**	**Total**
Substitution: manner of articulation	2,4,9,15,18,19,20,22,31,34,35,43,61,67,74,78, 89,109, 115,116,131,143,145,163	24
Substitution: place of articulation	4,9,15,19,20,22,34,43,61,74,75,78,89,106,109, 115,122,143,145,163	20
Substitution: voicing	15,19,31,34,35,43,61,63,67,74,109,122,127	13
Substitution (nature unspecified)	131	1
Omission	15,19,22,34,59,75,78,109,131,163	10
Excessive aspiration	35,48,74	3
Weak aspiration	109, 115, 116	3
Elongation	43,67,75	3
Addition	9,61	2
Undershoot	20	1
Tensing	67	1
Lenition	127	1

As demonstrated in **table 2.3**, consonants were especially affected by substitutions. Articulatory errors consisted of changes in manner, place and voicing of phonemes. Manner of articulation was mostly affected (*n*=24/59, 40.68%) [2, 4, 9, 15, 18, 19, 20, 22, 31, 34, 35, 43, 61, 67, 74, 78, 89, 109, 115, 116, 131, 143, 145, 163]. Noteworthy is the absence of flapping in four native English-speaking patients [18, 31, 67, 108]. Errors against all aspects (manner, place and voicing) were noted in seven cases [15, 19, 34, 43, 61, 74, 109]. Omission of consonants was attested in ten cases (*n*=10/59, 16.95%) [15, 19, 22, 34, 59, 75, 78, 109, 131, 163]. In comparison to vowels, consonants seem to be omitted more often than added.

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Table 2.4. Suprasegmental speech characteristics in patients with vascular FAS

SUPRASEGMENTAL		
Type	**Case**	**Total**
Slow(er) speech rate	9,18,19,20,22,24,31,34,43,74,89,96,109,112,115, 116,118,122,127,131,140,145	22
Altered rhythm (stress-timed speech, syllable-timed speech, ..)	18,19,21,22,24,38,43,44,48,49,67,74,75,78,109, 115,116,122,140,145	20
Abnormal intonation at end of sentence/utterance (F0 excursion on sentential level)	18*,20,21,31*,35,38,43,48,66,67,89,93**,**96,109, 115,116 ,131,163	18
Wrong word stress	2,17,18,24,31*,66,73,75,89,96	10
Long pauses	17,19,31,34,44,96,109,115,116	9
Altered melody	16,18,19,24,27,43,44,48,70	9
Dysprosody	4,24,61,131	4
Unexpected VOT - values	67*,127,140,163	4
Monotonous	109,115,116,145	4
Monopitch	24,89,96	3
Slow articulation rate	43,122,127	3
Scanned speech	131, 114	2
Excessive trill	116	1
Unaspirated syllable-initial plosives	40	1
Fast speech rate	78	1
Monoloudness	89****	1
Errors against assimilation	139	1
Nasalization	2	1

*18**= acoustically tested for spontaneous, read and repeated speech
*31**= spontaneous and read speech
*67** = longer lag VOTs for voiced plosives and shorter lag VOTs for unvoiced plosives
*89**= speech was also characterized by a 'flattened prosody'. This complicates interpretation, as prosody is not a monolithic concept. According to Kent (2004): 'Prosody consists of alterations in pitch, stress, and duration across words, phrases, and sentences. These same parameters are defined acoustically as fundamental frequency, intensity, and timing. It is the variation in these parameters that not only provides the melodic contour of speech, but also invests spoken language with linguistic and emotional meaning.' (*The MIT Encyclopedia of Communication Disorders*, p. 107)

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*93**= "She displayed pathologic [sic] patterns in prosody, vowel formant frequencies, vowel durations, and increased variability on these vowel measures, and she showed normal patterns in the production of voicing and place of articulation in stop consonants as well as preservation of the distinction between tense and lax vowels. Subsequent to her cerebellar stroke (CVA2), the patient's pathologic [sic] speech patterns receded and she showed normal speech output" (p. 566)

On a suprasegmental level, altered intonation, rhythm and word stress are the most striking features of FAS speech in vascular patients. As mentioned before, speech was found too slow in 22/59 stroke patients (37.29%) [9, 18, 19, 20, 22, 24, 31, 34, 43, 74, 89, 96, 109, 112, 115, 116, 118, 122, 127, 131, 140, 145]. Rhythm was off in 20 patients and this – when specified – almost exclusively referred to stress-timed languages becoming syllable-timed [18, 19, 21, 22, 24, 38, 43, 44, 48, 49, 67, 74, 75, 78, 109, 115, 116, 122, 140, 145] (33.90%). In 18/59 patients (30.51%) sentence intonation was aberrant [18, 20, 21, 31, 35, 38, 43, 48, 66, 67, 89, 93, 96, 109, 115, 116, 131, 163]. This almost exclusively referred to the use of a raised intonation at the end of the utterance (e.g. see Verhoeven et al., 2013; Kuschmann & Lowit, 2015: *'continuation rise'*), although in one case this also concerned a sudden unexpected fall in sentence intonation in a Persian patient [96].

2.3.7. Cognitive deficits associated with vascular FAS

In almost half of the vascular FAS cases, more specifically in 27 cases [4, 14, 17-19, 22, 24, 31, 38, 43, 59, 73-75, 78, 89, 108, 109, 111, 115, 116, 123, 125, 127, 131, 136, 143] (*n*=27/59; 45.76%), neurocognitive findings were reported. In 32 out of the 59 cases with vascular FAS (*n*=32/59; 54.24%) [1, 2, 7, 8, 13, 15, 16, 20, 21, 27, 28, 34, 44, 48, 49, 54, 55, 61, 66, 67, 70, 93, 96, 112, 118, 136, 139, 140, 145, 146, 149, 163] cognitive functions were not investigated. Cognitive impairments were identified in relatively few patients (*n*=9/59; 15.25%) [4, 31, 74, 78, 89, 115, 116, 125, 131].

Five out of these nine patients had some degree of acalculia [4, 74, 78, 115, 116]. For case 18 it was noted the patient had difficulty with more complex calculations. Acalculia is typically associated with frontal or parietal lobe damage (dominant angular gyrus). Case 78 had parietal damage and case 74 had damage affecting the frontal lobe, but for the remaining cases [4, 115, 116] the lesion location was not described in detail [case 115, 116: 'left hemisphere'; case 4: 'left MCA territory'] (see appendix A1). Case 4 and 78 had isolated deficits in arithmetic. For case 4, who also displayed agraphia, it is possible that the patient also had a (partial) Gerstmann syndrome.

Depressed working memory was noted in six patients [31, 89, 115, 116, 125, 131]. For case 131, both digit and visual span were affected (tested at 1 month and 6 months post-onset), which revealed a severe and sustained working memory deficit. For the cases 115 and 116, no information was provided as to which tests were employed.

For cases 89 and 131 executive dysfunctions were mentioned, but test results were

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only provided for case 131. Case 131, who suffered an extensive basal ganglia hemorrhage, had impairments in attention (digit span forward: pc. 2.3; visual span forward: pc. 7.5), executive functions, memory and non-verbal intelligence (digit span backward: pc. 0.5; visual span backward: pc. 7.5; verbal learning test: 1st trial: pc. 0.5 - 5th trial: pc. 0.5; Rey Complex Figure: immediate recall: pc. 1; delayed recall: 0.4; TMT A: pc. 0.5 and TMT B: 0.5; WCST: pc. 35; RCPM: pc. 4). Visuoconstructive abilities were spared (RCF copy). Cognitive deficits remained even after cognitive therapy (twice/week, 1 year; retested 12 months post-onset). Case 31 was diagnosed with a frontal lobe disorder following weak performance on the Luria 3-step test (Luria, 1970; 1980) (no scores available).

As reflected by pathological scores on all of the cognitive tests administered one month post-stroke (Mini Mental State Examination (MMSA; Folstein et al., 1975), Wechsler Adult Intelligence Scale-III (WAIS-III; Wechsler, 1998), Repeatable Battery for Assessment of Neuropsychological Status (RBANS; Randolph et al., 1998), Raven Progressive Colored Matrices (RCPM; Raven, 1976), Wisconsin Card Sorting Test (WCST; Grant et al., 1993), Trail Making Test (TMT; Reitan, 1958), the Middelheim Frontality Scale (De Deyn et al., 2005) and Frontal Assessment Battery (Dubois et al., 2000), a general cognitive decline in the context of Schmahmann's syndrome (Manto & Mariën, 2015) was found in case 125 following a right cerebellar hemorrhage. The scores on the executive tasks had deteriorated when retested 6 months post-stroke (Stroop task; Golden et al., 1978; WCST, and TMT).

2.3.8. Remission of FAS

Analysis of the entire study corpus revealed that the accent receded in only 23.21% of the patients (*n*=26/112) within the period of follow-up [9, 16, 24, 27, 41, 43, 45, 48, 53, 56, 60, 62, 78, 82, 83, 93, 98, 104, 108, 123, 124, 138, 145, 155, 156, 163, 169]. For vascular FAS, remission of FAS took place in 11 out of the 59 cases (18.64%) [16, 24, 27, 43, 48, 78, 93, 108, 123, 145, 163] (range: 1 day - 3 years). In seven of these patients the lesion was situated in the frontal lobe, affecting the precentral area and frontal gyri [16, 24, 43, 48, 93, 108, 163]. Three patients who had basal ganglia involvement recovered from FAS [27, 78, 123]. For case 145, the authors withheld (a) pontine lesion(s). In 25.58% (*n*=22/86) of the neurogenic patient group FAS receded. By contrast, seven out of the 18 patients (38.89%) with psychogenic FAS recovered from the condition during the follow-up period [41, 62, 82, 83, 98, 124, 169] (see also Keulen et al., 2016d).

Multiple chi-square analyses were conducted in order to answer: 1) whether the prognosis for patients suffering from psychogenic FAS is better than for those suffering from (acquired) neurogenic FAS, 2) whether there is a gender-related advantage in recovery and 3) whether there is an influence of the presence of comorbid speech and/or language disorders on the remission of FAS symptoms. For the purpose of these analyses, only patients with acquired neurogenic FAS were compared to those with psychogenic FAS, as groups would be too small if mixed FAS, developmental FAS and mechanic-orga-

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nic FAS were included. This resulted in a total study group of *n*=104 cases, of which 18 patients had psychogenic FAS.

For the first question, chi-square analysis demonstrated that the number of neurogenic patients (*n*=86) who recovered was not significantly different from the number of psychogenic patients (*n*=18). Hence, there was no significant relation between subtype and remission of FAS $X^2(1, n=104)=0.615,\ p=.433$).

Chi-square analysis for the second question revealed that the proportion of men (*n*=33) who recovered from FAS was not significantly different from the proportion of women (*n*=71). There was no significant relation between gender and remission of FAS $X^2(1, n=104)=1.37, p=.242$). This analysis was not replicated for stroke cases only, due to weak statistical power.

Lastly, for the chi-square analysis performed in response to the third question, cases with only pre-FAS mutism were excluded: only aphasia, apraxia of speech, dysarthria, agraphia, alexia, and agrammatism (in context of aphasia or not) were taken into account. Results revealed that there was no significant relationship between the presence of comorbid disorders and prognosis (remission) ($X^2(1, n=104)=.653\ p=.419$). Alternatively, there was also no significant relation between the occurrence of pre-FAS muteness and remission ($X^2(1, n=104)=.043\ p=.835$). Again, these analyses were not replicated for stroke cases only, due to weak statistical power.

2.4. DISCUSSION

2.4.1. Demographic findings and lesion location

A first finding of the present review of FAS cases published between 1907 and October 2016 shows that FAS occurs approximately twice as often in women (*n*=75/112), as in men (*n*=37/112). The difference between mean age for women and men in the corpus and stroke group was not significant. Nevertheless it is important to note the mean age of FAS-onset in the stroke population (51.57 years, range= 24-76 years, M= 53, SD= 12.51 years) is lower compared to other neurogenic motor speech disorders: Flowers et al. (2013) investigated mean age of first-ever acute stroke patients and found that the mean age of patients with dysarthria was 69.1 years (SD=14; *n*=92). For apraxia of speech, Ogar et al. (2006) obtained a mean age of 63.1 (SD=13.3; range: 31-79; *n*=18) years for stroke patients in the late phase of stroke (+1 year post-stroke). Hickok et al. (2014) included 17 patients with AoS in a study on the relation between AoS and verbal short-term memory. The patients, whom all suffered an acute ischemic stroke, were investigated in the acute stage and suffered a comorbid aphasia. Their study population had a mean age of 57.8 years (SD= 14.2).

Many of the stroke patients suffered comorbid speech and language disorders such as aphasia in the acute phase (33.90%, mean age: 47.5 years, SD: 13.89; *n*=20), in the lesion phase (3.39%, mean age: 35.5 years, SD: 13.44; *n*=2) or late phase: 1.69% (1.69%,

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age: 37; *n*=1), acute dysarthria (20.34%, mean age: 50.08 years, SD: 14.65; *n*=12), dysarthria in the lesion phase (5.08%, mean age: 46 years, SD: 10.82; *n*=3) and AoS in the acute stage (10.17%, mean age: 40.33 years, SD: 13.09; *n*=6).

The majority of FAS patients were right-handed: 52.68% (*n*=59/112). Crossed FAS in right-handed patients has been exceptionally reported in four cases: three stroke cases [8, 38, 70] and one case in which the lesion was of traumatic nature [23] (**table 2.1**). The incidence of crossed stroke-based FAS (*n*=3/59; 5.08%) between 1907 and 2016 is only slightly higher than the incidence of crossed aphasia. This is variably estimated between 0.38% and 4% in the stroke population (Hécaen et al., 1971; Carr et al., 1981; Mariën et al., 2004).

The vast majority of the vascular patients developed FAS after an isolated *left* hemisphere stroke (*n*=46/59; 77.97%), and in 6.78% of the stroke cases, lesions affected both hemispheres (*n*=4/59; 6.78%). The slight differences might be the result of the fact that handedness was only exceptionally investigated by formal instruments in 11.61% of the cases (*n*=13/112).

Although many authors have argued that AoS and FAS may be related disorders, anterior insular damage the neuroanatomical locus for AoS (Dronkers, 1996) – was not identified as an exclusive or even crucial neuroanatomical seat of vascular FAS. Evaluation of the study corpus shows that only 11.86% of the stroke cases had damage involving the left insula (*n*=7/59; 11.86%). Dronkers' (1996) hypothesis that the insula is the cardinal substrate for AoS has been challenged (see also Pyun et al., 2013). Hillis et al. (2004) found an association between damage of the left posterior inferior frontal gyrus (Broca's area), damage to the precentral and postcentral region, and AoS. Perfusion-weighted imaging demonstrated that even a hypoperfusion in Broca's area might be sufficient to evoke AoS in some patients. In 2014, Graff-Radford et al. pinpointed the premotor cortex and precentral gyrus as the crucial neuroanatomical substrate of AoS. They compared lesion locations between patients with AoS with and without comorbid aphasia, and looked for a shared area of structural vascular damage. Their findings correspond with the findings for patients in the current study corpus: cases 43 and 93 demonstrated simultaneous stroke-based AoS and FAS in the absence of aphasia. Case 136, who demonstrated FAS, AoS and aphasia, suffered an ischemic lesion in the area of the left insula, but also showed a hypoperfusion in the left motor area on SPECT. When only considering FAS, precentral sulcus and/or premotor involvement were present in 20.34% (*n*=12/59) of the vascular cases.

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However, within the group of vascular FAS patients, the majority of the patients had lesions that can (specifically) be situated near areas of importance in the cortical-subcortical connections that subserve motor speech behavior: some patients had lesions in or near Broca's area, affecting the inferior frontal gyrus [19, 43, 127, 163], the SMA, premotor or motor strip [16, 17, 22, 28, 43, 48, 74, 93, 108, 122, 143, 163], the insula [43, 48, 74, 78, 111, 127, 136], the left center semiovale [27, 96], the internal capsule with basal ganglia involvement [20, 31, 53, 55, 73, 78, 89, 123], or the basal ganglia [54, 59, 75, 125, 131, 143]. Others had a lesion in the fronto-temporo-parietal lobe [112], temporal lobe [67, 111], left parietal lobe [34], or fronto-parietal lobes [93, 122]. The (pre) motor area, SMA, and Broca's area fulfill an important role in planning, programming and integrating motor speech sequences. The basal ganglia and cerebellar regions, on their part, form the motor speech control circuits with frontal connections for (corrective) feedback (Duffy, 2013). The cortical (Brownsett et al., 2010; Tremblay et al. 2003) and subcortical (Catani et al., 2005) parts of parietal lobe fulfill an important role in converting sensory information into motor representations, but also in monitoring speech production (somatosensory cortex) (Tremblay et al., 2003).

Some vascular FAS patients had lesions largely confined to areas that at first sight seem less *directly* relatable to speech production disorders e.g. [14, 44, 109, 125, 139]. Case 44, for example, suffered an ischemic infarction in the body of the corpus callosum. The FAS in this patient was especially marked by prosodic deficits, which the authors defined as a "linguistic aprosody" (p. 1551). Monrad-Krohn (1947) also termed FAS in his patient as a *dysprosody*, which in his description refers much more to 'linguistic prosody'. His patient's musical prosodic abilities were unaffected and prosodic-linguistic problems were situated at the level of word accent and sentence intonation. He furthermore qualified the patient's condition as "an 'ataxia' of the prosodic faculty" (p. 411). Whitaker (1982) considered the typical timing and target errors of the patient he reported on as analogous to ataxic dysarthria and speculated that the cerebellum might be functionally implicated via anatomical connections with the inferior rolandic cortex which, he argued, acts as a pivotal center in the speech production system.

2.4.2. Linguistic characteristics of vascular FAS and comorbid speech and language deficits

As shown in **table 2.5**, the semiological similarities between FAS, AoS and ataxic dysarthria are striking. The three disorders are difficult to dissociate, especially on the segmental level. AoS and ataxic dysarthria are generally associated with vowel and consonant distortions, although for AoS, consonants have been argued to be attained more, whereas for ataxic dysarthria especially vowel articulation is affected. One of the major dissociations between FAS and AoS is situated at the level of fluency: the trial-and-error behavior (groping), the false articulatory starts and syllable repetitions are absent in FAS speakers (Duffy, 2013). How can the clinical resemblances be explained?

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First, for AoS and FAS specifically, there is the cognitive explanation as proposed by Whiteside & Varley (1998). These authors argued that the characteristics of both AoS and FAS are the result of compensatory strategies for defective access to, or retrieval of verbo-motor patterns. They argued that AoS and FAS should be considered as two poles of a 'severity'-continuum, with FAS representing the mildest form of a speech planning disorder, in which the patient preserves the most efficient – although not perfect – compensation techniques, and AoS at the opposite pole, representing the most severe speech planning impairment. Compensation is explained in terms of direct and indirect speech encoding routes: the indirect route requires on-line speech encoding. This is expected to occur more often in low frequency words, possibly with a high phonetic complexity. The direct route relies on instant retrieval from storage. This is argued to be the case for high frequency words, and demands a lower cognitive load. Due to disruption to the direct route, the indirect route will be called on more often in AoS and FAS, but encoding is more efficient in case of the latter impairment.

In other cases, FAS has been explained in the context of the Directions In Velocity of Articulators or DIVA- model (Guenther, 1994), which allows for the explanation of MSD's in the context of (failed) feedback and/or feedforward control processes. However, the variable lesion locations in the context of FAS are problematic, as they do not allow for the designation of a clear-cut impairment of a particular structure in the model. Tomasino et al. (2011) situate the deficits in FAS at the level of feedforward control. Their patient was a 50-year-old, right-handed, monolingual Italian-speaking woman suffering a tumor encompassing the precentral gyrus (motor strip, BA 4). This lesion would entail disrupted feedforward control, whereas FAS in cases with primarily subcortical lesions (e.g. basal ganglia lesions) would be associated with feedback control. Information about the speech sound maps (localized near inferior frontal gyrus and parts of the premotor cortex) are sent to articulator and velocity maps in the precentral gyrus. In their patient, the precentral gyrus was infested by a tumor. They argue that as a consequence of this, the integration of the commands for articulator and velocity maps, failed and could as such explain the FAS.

In agreement with AoS characteristics (**table 2.5**), FAS is often characterized by a very slow speech rate (*n*=22/59; 37.29%). The relative frequency of this finding might be explicable in association with other, related characteristics. For instance, vowel lengthening occurred in 15 patients (*n*=15/59; 25.42%) and was directly associated with slow speech in 8/15 patients.

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Table 2.5. Overview of the segmental and suprasegmental characteristics of apraxia of speech and ataxic dysarthria.

APRAXIA OF SPEECH		ATAXIC DYSARTHRIA	
SEGMENTAL		**SEGMENTAL**	
Voicing errors	Freeman (1987), Itoh & Sasnima, (1984); Itoh et al., (1982), Kent & Rosenbek, (1983); Ziegler, (1987), Dabul et al. (2000), Duffy (2013)	Segment lenghtening	Hartelius et al. (2000), Duffy (2013), Darley et al., (1969a)
Place of articulation	Duffy (2013)	Errors of articulatory movement	Hartelius et al (2000)
Manner of articulation	Duffy (2013), Ogar et al., (2007)	Errors of articulatory range	Hartelius et al. (2000)
Vowel errors	Dabul et al. (2000), Duffy (2013)	Distorted vowels	Darley et al. (1969a), Duffy (2013)
Inconsistent errors	Dabul et al. (2000)	More vowel errors than consonant errors	Duffy (2013)
Addition (schwa)	Darley et al. (1975), Dabul et al. (2000), McNeil et al. (2004)		
Vowel elongation	Hardcastle (1987), Ballard et al. (2000), McNeil et al. (2004), Duffy (2013)		
Consonant elongation	McNeil et al. (2004)		
Reduced co-articulation	Mayer (1995); McNeil, et al. (1994); Ziegler & von Cramon (1985, 1986b)		
'Segment' prolongation	Kent & Rosenbek (1983)		
Deletion (cluster reduction)	Ballard et al. (2000), Duffy (2013), Ogar et al., (2007)		
Perseveration errors	Dabul et al. (2000)		
Anticipation errors	Dabul et al. (2000)		
Transposition errors	Dabul et al. (2000)		
Initiation difficulty	Dabul et al. (2000)		
More errors against consonants than vowels	Darley et al. (1975)		

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SUPRASEGMENTAL		SUPRASEGMENTAL (incl. voice quality)	
Abnormal intonation	Wertz et al. (1984), Ogar et al. (2005)	Monopitch	Darley et al. (1969 a,b), Kent et al. (2000), Hartelius et al. (2000), Duffy (2013)
Altered rhythm	Wertz et al. (1984), Ogar et al. (2005), Duffy (2013)	Monoloudness	Darley et al. (1969 a,b), Kent et al. (2000), Duffy (2013)
		Harshness (possibly transient)	Darley et al. (1969 a,b), Hertrich et al. (1998), Duffy (2013)
Abnormal prosody	Dabul et al. (2000)	Pitch breaks	Joanette & Dudley (1980)
Slow speech rate	Kent & Rosenbek (1983), McNeil et al. (2004), Ogar et al. (2005), Duffy (2013)	Altered pitch level	Joanette & Dudley (1980)
Equal stress	McNeil et al. (2004), Duffy (2013), Ogar et al., (2007)	Alternating loudness	Gilman & Kluin (1992), Hartelius et al. (2000), Duffy (2013)
Unusual accent	McNeil et al. (2004), Duffy (2013)	Fluctuating pitch	Gilman & Kluin (1992), Hertrich et al. (1998), Hartelius et al. (2000)
Prolonged intersegment durations (Pauses)	Ballard et al. (2000), McNeil et al. (2004), Duffy (2013), Ogar et al., (2007)	Breathiness (transient)	Gilman & Kluin (1992), Hertrich et al. (1998)
		Voice tremors	Gilman & Kluin (1992), Hertrich et al. (1998), Duffy (2013)
		Irregular intersegment durations	Ackermann & Hertrich (1994), Boutsen et al. (1997), Gentil et al. (1990 a)
		Regular intersegment durations	Hartelius et al. (2000)
		Altered rhythm (often stress times --> equalization of syllables)	Ackermann & Hertrich (1994), Hartelius et al. (2000), Kent et al. (2000)
		Scanned speech	Kent et al. (2000), Duffy (2013)
		Altered stress patterns	Kent et al. (2000), Duffy (2013)
		Flat F_0	Hartelius et al. (2000)
		Slow speech rate	Hartelius et al. (2000), Duffy (2013)

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Epenthesis, addition and diphthongization can also directly and indirectly influence articulation and speech rate and the tendency to raise the tongue could be at play in creating the impression of a longer vowel duration: raising often concerns increasing tenseness in both front (15, 17, 38; esp. /I/) and back vowels (17, 89, 109; esp. /a/ and /ɔ/).

Dysarthria occurred at approximately the same time as FAS in 12 out of the 15 vascular FAS cases who presented with comorbid dysarthria: in 9 cases they both occurred in the acute phase [14, 17, 20, 22, 27, 89, 96, 125, 136], in 3 cases they both occurred in the lesion phase [16, 38, 131]. In two cases, dysarthria preceded FAS [31, 49]. For case 13, the speech of the patient was initially dysarthric, however it was not possible to situate the onset of FAS in relation to stroke onset. For most patients, the speech characteristics associated with FAS corresponded with what can be seen in dysarthric patients (irrespective of the type) [17, 20, 22, 27, 31, 38, 49, 89, 96, 125, 131] (see **table 2.2-2.5**).

The muscular tension hypothesis forwarded by Graff-Radford et al. (1986) and Ingram et al. (1992) draws a relation between FAS and dysarthria. Graff-Radford et al. (1986) described a 56-year-old right-handed American English-speaking woman who suffered a stroke in the left frontal lobe, extending to the basal ganglia. The patient's segmental changes (primarily raising, but also backing and lowering of vowels), which gave rise to a Nordic accent, were attributed to increased muscle tension. This hypothesis was based on perceived segmental changes. Ingram et al. (1992) re-evaluated this hypothesis on the basis of acoustic data and refined it: the raised muscular tension and associated excursions of articulators from neutral position when articulating segments could explain part of the segmental changes. Ingram et al.'s (1992) patient demonstrated consonantal fortition, and less weakening processes. The researchers argued that FAS characteristics are less stable than in other executive MSD's, such as dysarthria. Although they did not offer an explanation for the suprasegmental changes, they posited that altered segmental structures (e.g. less weakening, less vowel reduction) have inevitable effects on the suprasegmental level. This stands in stark contrast to the hypothesis forwarded by Monrad-Krohn (1947) (see also Blumstein et al. 1987) who argued that the segmental errors are secondary to the prosodic disturbance.

Related to these theories is the fortition hypothesis adhered to by van der Scheer et al. (2013) (see also Jonkers et al., 2016). Fortition stands in direct relation to an increased muscular tension. Verhoeven & Mariën (2007) however, provided an alternative explanation for the occurrence of this phenomenon, arguing that fortition may be the result of relying more heavily on proprioceptive feedback, and consequently opting for the realizations that give more clearly defined feedback (i.e. stops). The cerebellum and basal ganglia have been argued to play an important role in proprioception. Lesion studies have demonstrated that the cerebellum allows for the realization of predictive models that enhance proprioception (e.g. Boisgontier & Swinnen, 2014).

Interestingly, in the early days, AoS was also referred to as ataxic aphasia or cortical dysarthria, already hinting at a semiological resemblance, and by inference, at a specific role for the cerebellum in this speech disorder (Kertesz, 1984). From a semiological

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standpoint, AoS and FAS are remarkably similar to the speech disturbances consequent to lesions involving the superior paravermal region of the cerebellum, i.e. ataxic dysarthria (Kertesz, 1983; Ackermann et al., 1992; Mariën et al., 2001, Mariën et al., 2006). A slow and strikingly irregular articulation, a monotonous, staccato and scanned oral-verbal output, inconsistent misarticulations of vowels and consonants, and deficits affecting voice onset time (the time interval between consonant burst and vowel onset) and production and discrimination of vowel length are commonly found in AoS, FAS and ataxic dysarthria (Mariën et al., 2001; Ackermann & Hertrich, 2000) (see also **Table 2.5** above).

From a pathophysiological point of view, functional neuroimaging data has demonstrated that in contrast to the long-standing belief that FAS and AoS are solely produced by lesions of the anterior perisylvian speech areas, the cerebellum may be crucially involved in motor speech planning disorders (Mariën et al., 2006; Mariën & Verhoeven, 2007). In addition to a substantial amount of clinical and experimental evidence in support of a functionally lateralized linguistic cerebellum (Mariën et al., 2001 a,b) a number of cases – including ones in the current corpus – indicate that the posterior fossa structures are implicated in FAS [75, 85, 125; see also Keulen et al., accepted)], although these structures are usually more directly associated with *executive* motor speech disorders of the dysarthric type and/or mutism (e.g. Frim & Ogilvy, 1995; Kumral et al., 2002).

The timing and coordination functions subserved by the cerebellum have a direct effect on the suprasegmental features e.g. syllable-timed speech, excessive pausing, staccato rhythm, and slow rate (usually associated with ataxic dysarthria), but can also explain the segmental distortions, as the cerebellum plays a cardinal role in regulating timing and force of muscular movements to achieve targets. The cortex projects information concerning movement schemes or plans to the cerebellum, which integrates the somatosensory information to make predictions in order to reach targets (its proprioceptive function). Connections with the basal ganglia allow for corrective feedback when required (e.g. Ackermann & Hertrich, 2000; Ackermann, 2008; Boisgontier & Swinnen, 2014).

Based on the suprasegmental and segmental findings for the current corpus, we argue FAS is a dual component motor speech disorder with a concurrence of articulatory planning and executive deficits; explaining the shared characteristics with ataxic dysarthria and AoS. Interestingly, Scott et al. (2006) already argued that FAS may be the result of "a disconnection of the planning of articulation from motor control" (p. 370). Their patient was a 36-year-old right-handed Scottish woman who suffered a stroke in the left hemisphere in the white matter near the precentral sulcus and anterior insula and developed a foreign accent described as German, Polish, or South African. Scott et al. (2006) stated that the role of the insula encompasses the integration of phonological features with suprasegmental features. The disruption of this process would entail FAS-like speech and can be seen as the consequence of a disruption between the motor strip and

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anterior insula, which means that they too were possibly regarding FAS as a disorder of at once speech planning (such as AoS) and execution (such as dysarthria).

Finally, FAS has equally been qualified as an '*aphasic* epiphenomenon' (Ardila et al., 1988). According to Ardila et al. (1988) "loss of verbal fluency", "broadening of phonemic boundaries (a greater phonetic dispersion which can lead to the appearance of phonological paraphasias)", "inadequate use of suprasegmental features of language" and "a certain degree of agrammatism" (p. 497) enhance the impression of a foreign-accented speaker. They argue that previous symptoms are typically associated with (non-fluent) aphasia.

In a related way, Critchley (1962) also distinguished FAS as a sort of post-aphasic dysprosodia (see also Monrad-Krohn, 1947). However, aphasia preceded the onset of FAS in only five cases (of vascular FAS) [2, 16, 31, 49, 112]. According to Varley et al. (2006) "mild aphasic impairments such as hesitation associated with lexical retrieval difficulty and syntactic and morphological errors contribute to the perception of 'foreignness'" (p. 357). In contrast, Kurowksi et al. (1996) reject any relation to Broca's aphasia, mainly referring to the flattened intonation typifying non-fluent aphasic patients' speech. They argue that FAS is 'qualitatively distinct' from Broca's aphasia. It is interesting to note that for three stroke patients, lesion location was explicitly stated to comprise Broca's area (posterior part of the inferior frontal gyrus) [19, 43, 163].

2.4.3. Cognitive deficits in vascular FAS

Stroke is usually associated with some degree of cognitive impairment. Patel et al. (2002) variably estimated the prevalence of cognitive deficits in the stroke population between 11.6% and 56.3% and calculated that 38% of the stroke patients included in their study (*n*=645) were cognitively impaired at three months post-stroke (MMSE scores: <24/30). In post-stroke aphasia, cognitive deficits are common, and a relationship has been found between aphasia-severity and cognitive deficit (Lee & Pyun, 2014).

This review shows that cognitive deficits were not systematically looked for. Cognitive impairments were only identified in nine out of the 59 stroke patients [*n*=9/59; 15.25%]. Seven of these patients presented with comorbid speech and language disorders. The majority of these patients had lesions situated within the frontoparietal areas [4, 31, 74, 78, 89], for cases 115 and 116 the lesions were not clearly circumscribed and for cases 125 and 131 they were situated in respectively the cerebellum and basal ganglia.

Seven patients had problems with arithmetic [4, 31, 74, 78, 115, 116, 125]. For case 4, acalculia was the only cognitive impairment. Case 115 and 116 suffered from short-term memory problems as well as deficits in arithmetic, although no test results were indicated in the report. Case 74 did not demonstrate any major cognitive deficits. Although general intelligence was normal (VIQ= 94; PIQ= 98), the authors report on 'low' scores for the arithmetic and picture completion task, without providing the exact

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scores. On the delayed memory (recognition) task she underperformed for verbal material (14/25) and performed below her age norm for visual material (39/50). Performance on the Weigl Colour-Form sorting test, a non-verbal test investigating set-shifting, was normal (no scores were provided). Case 78 had deficits with respect to arithmetic and a poor general mental status (MMSE: 22/30). By contrast, full scale IQ (117), visuospatial abilities (ROCF; judgment of line orientation task), complex visual recognition (visual form discrimination task) and left-right orientation were normal.

Arithmetic requires intact short- and long-term memory functions, attention, and mental flexibility (further impaired in case 74, 115, 116; Alexander et al., 1986) and is subserved by mainly parietal and (pre)frontal cortices (see Rosenberg-Lee et al. 2011; Menon et al., 2000; Menon et al., 2002). Basal ganglia lesions, attested for cases 31, 78, 89, and 127, give rise to similar cognitive impairments as frontal lesions due to a network of fronto-subcortical loops (Casey et al., 2002, Delazer et al., 2004). The basal ganglia are implicated in a wide variety of cognitive functions, including executive functions, attention, visual perception, sequential processing and learning – which were variably disrupted in patients 31, 74, 78, 89, 125, 127 – but also speech (Brown et al., 1997). These behavioral effects are demonstrated in vascular FAS cases with basal ganglia damage [31, 89, 127].

Interestingly, Reeves & Norton (2001), Blumstein & Kurowski (2006) and Schiff et al. (1983) related FAS to disruption of the cortico-striatopallidal-thalamic pathway, consisting of many regions that cooperate in an integrated fashion to regulate many different cognitive functions and though there are still many issues to be resolved, the circuit is clearly involved in various speech and language processes (production and control) (Groenewegen et al., 1990). However, in this account the cerebellum was not included. Cerebellar lesions affecting cerebro-cerebellar connections, which involve the basal ganglia, can give rise to cerebro-cerebellar diaschisis and induce frontal-like cognitive and behavioral deficits due to functional disconnection with regions crucially implicated in higher cognitive functions, but also in motor speech control (see Leiner et al., 1986, 1989, 1991, 1993; Middleton et al., 2000; Bailleux et al. 2008, 2010; Stoodley & Schmahmann, 2010). The most poignant example of the distant functional effect of cerebellar pathology is case 125. Following a right cerebellar hemorrhagic stroke, neurocognitive testing in this patient revealed persistent deficits on a range of tasks[6]. Due to a constellation of executive, spatial, affective and linguistic deficits the patient was diagnosed with cerebellar cognitive affective syndrome (CCAS) or Schmahmann's syndrome (Schmahmann & Sherman, 1998; Manto & Mariën, 2015).

2.4.4. Remission of FAS

In terms of remission, it was found that FAS receded during follow-up in only 23.21% of the patients. When taking into account only stroke cases, the figure decreases to

[6] For an overview of the results we refer the interested reader to Mariën et al. (2013)

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18.64%. For the stroke cases for whom remission was noted, FAS duration ranged from 1 day [108] to approximately 3 years [93, 123]. For the cases with FAS resolution, all but three [108, 123, 145] had comorbid speech disorders [16, 24, 27, 43, 48, 78, 93, 163]. None of them received a therapy specifically targeting the FAS.

Positive prognosis (remission as opposed to no remission) could not be associated with subtype, nor with the presence or absence of comorbid disorders based on the statistical analyses. This may have to do with weak statistical power due to the small number of available reports and data, especially when subdivided according to subtype. Hence, we stress the importance and the need for publication of more patient reports.

2.5. THE MANY PROBLEMS OF WHITAKER'S CRITERIA

Although the criteria proposed by Whitaker (1982) are still widely applied in the FAS literature, they substantially narrowed the original concept of a change of accented speech by restricting the condition to: 1) monolinguals, with 2) acute CNS damage whose oral verbal output is 3) subjectively considered foreign on a pure perceptual basis by the patient himself, by acquaintances or by the investigator. Reviewing the literature on FAS and its related topics since the first description at the beginning of the 20th century we found 27 cases that met all Whitaker's criteria for a FAS diagnosis [4, 15, 18, 19, 20, 59, 70, 73, 78, 89, 91, 104, 109, 115, 116, 118, 131, 138-140, 143, 144, 146, 148-150, 163]. This group of cases represents 24.11% (*n*=27/112) of the patients in our study corpus. How can it be that this group is so small?

The ear of the beholder: the golden standard?

First, in Whitaker's definition the diagnosis of FAS strongly depends on the *subjective* perceptual interpretation of the speech qualities by the patient himself, by acquaintances or by the investigator. As a result, much of the variability of perceived accents in perceptual FAS experiments follows from the lack of objective phonetic measures to semiologically identify FAS as a coherent and distinct motor speech disorder. As such, the diagnosis of FAS fundamentally suffers from subjective interpretation that crucially relies on the judgment of speech qualities based on the strongly variable implicit saliency of segmental and suprasegmental speech features in interlocutors. As a result, FAS has been argued to originate in the ear of the beholder (Kurowski et al., 1996).

Etiological differentiation

Second, several cases have been reported in which FAS did not clearly relate to central nervous system damage, or to an acute cerebral insult. Indeed, similar to Critchley's (1962; 1970) case 1, several other patients (*n*=24/112; 21.43%) [6, 41, 62, 63, 82, 83, 87, 90, 94, 95, 97, 98, 110, 124, 129, 130, 141, 142, 157, 165, 168, 169, 170, 171] have been

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reported who developed FAS without demonstrable brain lesions on structural brain imaging with CT or MRI. Most of these fit the diagnosis of a psychogenic FAS (*n*=18/112; 16.07%) [6, 41, 62, 63, 82, 83, 90, 97, 98, 110, 124, 129, 130, 157, 165, 169, 170, 171] (see Keulen et al., 2016d for a review).

A fourth FAS variant?

Cases 141 and 142, described by Dilollo et al. (2014) do not fit any of the neurogenic, psychogenic or mixed subtypes of FAS (Verhoeven & Mariën, 2010a). The authors described an accent change after a temporo-mandibular-joint surgery. An acute MRI of the brain was inconclusive for both patients due to artifacts from surgery (metal braces and implants), although case [142] underwent an MRI 6 months post-surgery, which demonstrated no structural lesions (see above). Both patients had always spoken with an American accent prior to surgery. Post-surgery, case 141 developed an Irish accent and case 142 spoke with a British English accent. For case 141 the accent diminished after approximately 18 months. Interestingly, psychological changes are pointed out for case 142, but these were in fact induced *after* the onset of the accent. After surgery, the patient even travelled to Great Britain, "in search of someone who sounded like [she did]" (p. 24). She described the psychological changes – of which she was fully aware – as positive. The patient embraced the accent and admitted it turned her into a more open, out-going person. The accent maintenance in case 142 could have been reinforced by the psychological changes affecting her person. This reinforcement of accent, which has been noted for other FAS cases in the past (e.g. Tailby et al., 2013), could be interpreted as an epiphenomenon that can affect all FAS patients, irrespective of the subtype. However, the question remains as to what explains the foreign accent after TMJ surgery. Although the authors did not investigate the speech of the patients, we hypothesize that the changes are dysarthria-like, induced by the manipulation of mechanics of the oral cavity. Because these cases did not develop FAS after neurological damage, but rather after structural changes to the speech organs, we propose to categorize these cases under a new, fourth FAS subtype: the organic-mechanic FAS.

Reversion to a previously learned accent.

Furthermore, as stipulated by the last of Whitaker's criteria, the delineation of FAS to patients with no evidence in their background of being a speaker of a foreign language artificially restricts the condition to rather small linguistic minorities of the world's population that stem from monolingual communities. This view on linguistic reality is evidently too narrow as most of the world's population is bi- or multilingual. Twenty-six out of the 112 cases (23.21%) were described as bilingual[7] [2, 7, 16, 27, 34, 67, 92, 98,

[7] The authors are aware that concepts such as 'bilingualism' and 'polyglot' can be defined in different ways. For the purpose of establishing this corpus all types (compact, coordinated, subordinated, balanced, dominant - Fabbro, 2003) were included, without making distinctions. We entirely followed the authors' descriptions.

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111, 125, 127, 136] or polyglot patients [13, 14, 31, 43, 46, 60, 63, 80, 94, 112, 145, 166, 167, 169]. For reasons of their bilingual or polyglot background, they do not survive Whitaker's criteria. However, as a change of accent and prosodic alterations might be an intrinsic symptom of pathological language mixing (Perecman, 1989), additional research is warranted to discriminate multilingual patients with FAS from patients who developed an accent in the context of bilingual or polyglot aphasia, which would exclude at least eight bilingual or polyglot vascular FAS cases, for whom aphasic symptoms were noted [2, 16, 31, 34, 67, 78, 112, 136].

In 28/112 cases (25%), it was clear that patients had had prior contact with the language associated with the newly acquired accent. In 13 of these patients (n=13/28), it was reported that the patients were in fact bilinguals or polyglots. The remaining patients had knowledge of the language(s) and associated accents due to intense language contact (with for instance relatives), or a basic level of formal education at school. Berthier et al. (2015) argued for the recognition of this disorder as a separate FAS variant. Indeed, in agreement with the hypotheses formulated by others such as Seliger et al. (1992), it could very well be that a disruption of a previously suppressed neural system leads to the surfacing of linguistic features the patient internalized at some point during linguistic development, but, for some reason, did not need and hence suppressed. Or, as Ojemann & Whitaker (1978) argued, that languages learned at different ages are localized in anatomically distinct regions in the brain. This hypothesis has been at the center of debate for many years now and other confounding variables have complicated the polemics: not only age of acquisition, but also type of instruction, and amount of language use play a role in the functional organization of the polyglot brain (Kim et al., 1997; Dehaene et al., 1997; Hernandez & Li, 2007; Perani et al., 1998; Perani et al., 2003; Perani & Abutalebi, 2005)

A need for revision

Reviewing Whitaker's set of criteria it seems that they do not provide a sufficiently reliable and solid basis for an unambiguous diagnosis of the clinical phenomenon in which motor speech alterations lead to a pseudo-accent or the subjective impression of a 'foreign accent' by interlocutors of the same speech community (Keulen et al., 2016b, Keulen et al., 2016c). We suggest to broaden the definition of FAS as a neurologically, psychiatric, mixed or mechanical induced motor speech planning and execution disorder in which segmental and/or suprasegmental changes lead to the impression of an altered accent by interlocutors of the same linguistic community.

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CHAPTER 3

The Posterior Fossa and Foreign Accent Syndrome: Report of Two New Cases and Review of the Literature[1]

[1] The study reported in current chapter was submitted in an adapted format as: Keulen, S., Mariën, P., van Dun, K., Bastiaanse, R., Manto, M., Verhoeven, J. (accepted). The posterior fossa and foreign accent syndrome: report of two new cases and review of the literature, *The Cerebellum*.

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ABSTRACT

Foreign accent syndrome is a rare motor speech disorder that causes patients to speak their language with a non-native accent. In the neurogenic condition, the disorder develops after lesions (often stroke or trauma) in the language dominant hemisphere, often affecting Broca's area, the insula, the supplementary motor area and the primary motor cortex. We present two new cases of FAS after posterior fossa lesions. The first case is a 44-year-old, right-handed, Dutch-speaking man who suffered motor speech disturbances and a left hemiplegia after a pontine infarction. Quantified SPECT showed a bilateral hypoperfusion in the inferior lateral prefrontal and medial inferior frontal regions as well as a significant left cerebellar hypoperfusion. Further clinical investigations led to an additional diagnosis of brainstem cognitive affective syndrome that closely relates to Schmahmann's syndrome. The second patient was a 72-year-old right-handed polyglot English man who suffered a stroke in the vascular territory of the left posterior inferior cerebellar artery (PICA) and developed a foreign accent in his mother tongue (English) and in a later learned language (Dutch). In this paper we discuss how the occurrence of this peculiar motor speech disorder can be related to a lesion affecting the posterior fossa structures.

3.1. INTRODUCTION

3.1.1. Foreign accent syndrome

During the last century more than 170 reports of patients with Foreign Accent Syndrome (FAS) have been published. This condition causes the patient with FAS to be perceived by speakers of the same language community as a non-native speaker of their mother tongue, due to speech changes at the segmental[8] and suprasegmental[9] level. FAS has been related to various etiologies including stroke, cerebral trauma, multiple sclerosis, dementia and psychiatric disorders. In 2010, Verhoeven & Mariën (Verhoeven & Mariën, 2010a) distinguished three different subtypes of FAS: neurogenic FAS (Verhoeven & Mariën, 2002), including developmental cases (Keulen et al., 2016a), but also psychogenic FAS (Keulen et al., 2016d) and a mixed-type FAS (Tailby et al., 2013).

The neurogenic variant is the most widely reported type. Although most of the patients with this subtype acquired FAS after stroke (Verhoeven & Mariën, 2002; Cole, 1971, case 2; Kwon & Kim, 2006; Naidoo et al., 2008; Mariën & Verhoeven, 2007, case 2) or brain trauma (e.g. Monrad-Krohn, 1947; Niels & McKeown, 1961, case 2; Edwards et al., 2005, case 4 & 5), the lesion sites do not unequivocally point towards a single neuro-

[8] the speech segments e.g. phonemes

[9] the contrastive aspects marking speech that are not identified as segments, but rather define the segments e.g. rhythm, intonation

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anatomical substrate. In the majority of the vascular cases, the causative lesions were located in the language dominant (left) hemisphere, especially in the frontal (e.g. Berthier et al., 1991, case 3; Denes et al., 1995, Munson & Heilman, 2005), fronto-temporal (e.g. Blumstein et al., 1987; Avila et al., 2004=González-Álvarez et al., 2003), and fronto-temporo-parietal regions (Monrad-Krohn, 1947; Levy et al., 2011; Masao et al., 2011). Although handedness was not formally investigated, 'crossed FAS' has been reported in four dextral patients after right-hemisphere damage (Critchley 1962, case 3; Berthier et al., 1991, case 2; Miller and O'Sullivan, 1997 = Miller et al., 2006; Dankovičová & Hunt, 2011). In three cases the cerebellum was functionally affected (hypoperfusion) after frontal lesions (mechanism of diaschisis) as evidenced by SPECT scans (Hwang et al., 2001; Verhoeven & Mariën, 2002; Mariën & Verhoeven, 2007, case 2). In six other cases, FAS was directly attributed to posterior fossa lesions (Cole 1971, case 1 & 2; Dankovičová & Hunt, 2011; Cohen et al., 2009; Tran & Mills, 2013; Mariën et al., 2013; see Appendix B1).

In this study we present a survey of the literature and two new cases of FAS after a posterior fossa lesion: the first patient suffered a stroke in the right pons and was described as an example of brainstem cognitive affective syndrome by Mariën & D'aes (2015). The second patient incurred FAS after a stroke in the left posterior cerebellum.

3.2. THE POSTERIOR FOSSA AND FAS: TWO NEW CASES

3.2.1. Case 1

A 44-year-old, right-handed, Dutch-speaking man with an educational level of 11 years was admitted to hospital on April 20th, 2012 after acute loss of consciousness followed by motor speech disturbances and a left hemiplegia. The patient had used 'poppers'; alkyl nitrites which, when aspirated, arouse euphoric feelings and relax sphincter muscles to enhance sexual experience. CT and electrocardiography on admission were normal. Five days later, the patient was transferred to the Neurology department of ZNA Middelheim Hospital. The neurological examinations on admission revealed a FAS and a paralytic dysarthria, a left facial nerve paralysis, a tongue protrusion to the left, a left hemiplegia, and a left hypertonia. The patient tested positive for (neurovascular) syphilis (blood and CSF analysis) and was subsequently treated with penicillin for two weeks. On admission, the medical staff had the impression that the patient spoke with a Hollandic-like accent. Speech was generally very slow, and had a pronounced nasal quality. Vowel pronunciation was affected by omissions (especially of schwa) and backing. In addition, diphthongs were often realized as monophthongs. Consonants were regularly devoiced (especially fricatives), or omitted (especially plosives - alveolar and velar). T1-weighted MR imaging of the brain demonstrated a hypointense lesion at the level of the right pons. T2-weighted axial FLAIR-imaging showed a hyperintense signal in the right pons (**Figure 3.1**).

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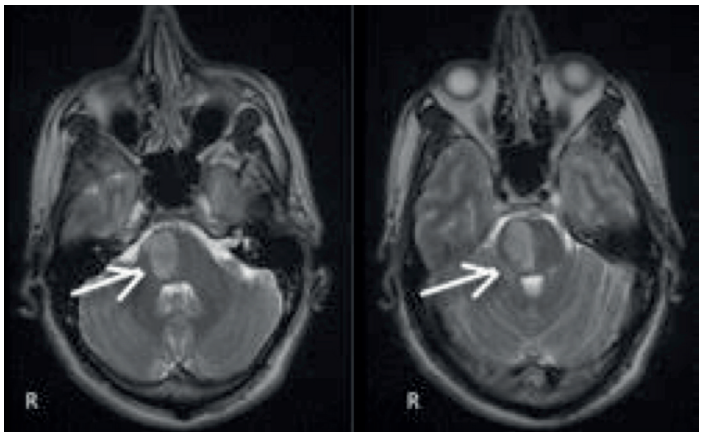
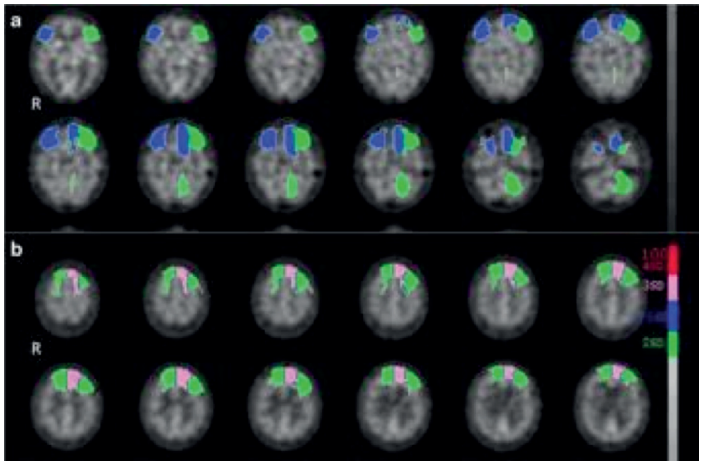
Figure 3.1. T2-weighted axial MRI slice of the brain demonstrating an ischemic lesion in the right pons (indicated by the white arrows).



A quantified 99mTc ECD SPECT scan in May 2012 showed a bilateral hypoperfusion in the inferior lateral prefrontal (left: -2.2 SD, right: -2.7 SD) and left medial inferior frontal regions (-2.8 SD). A significant left cerebellar hypoperfusion was equally retained (-2.1 SD) (**Figure 3.2**).

Figure 3.2. Quantified 99mTc ECD SPECT showing a bilateral hypoperfusion in the inferior lateral prefrontal and left medial inferior frontal regions as well as a significant left cerebellar hypoperfusion (a) and a significant bilateral frontal hypoperfusion (b).



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The EEG recording showed a mild impairment over both hemispheres: the polyrhythmic activity on the left showed a fast component whereas on the right a slowing of activities was found. Soon after admission the patient had a grand mal seizure and treatment with valproic acid was started. A transesophageal echocardiogram disclosed mitral insufficiency and a patent foramen ovale with a clear right to left shunt.

Extensive neuropsychological testing was performed in May 2012 (see **table 3.1**). Test results disclosed normal general cognitive abilities (MMSE: 27/30; -0.5 SD) (Folstein et al., 1975), a clinically deficient full scale IQ (FSIQ = 73; -1.8 SD) and a pathological performance IQ (PIQ = 61, -2.6 SD, with depressed scores for digit symbol substitution (-3 SD), block design (-1.3 SD), matrix reasoning (-1.7 SD), picture completion, (-1.7 SD) and picture arrangement: (-2.7 SD), whereas subtest scores of the verbal IQ were within the normal range (VIQ = 83; -1.1 SD) (Wechsler Adult Intelligence Scale-3rd Edition; Wechsler, 1998). The patient obtained pathological scores on the attention subtests (index 66, -2.3 SD) and delayed recall subtests (index 61, -2.6 SD) of the Wechsler Memory Scale – revised (WMS-R) (Wechsler, 1987). The visual memory index amounted to 70 and verbal memory to 80. Scores on the Trail Making Test were depressed (TMT A: <pc. 10; TMT B: <pc. 10) (Reitan, 1958). On the Wisconsin Card Sorting Test (Grant & Berg, 1993), the patient only completed 3 categories (-2.3 SD). A pathological score was found on the Stroop Color Word Test (card 1: < pc. 1; card 2: < pc. 1; card 3: pc. 3) (Stroop, 1935), and the Frontal Assessment Battery: 13/18 (Dubois et al., 2000).

In October 2012 the patient was re-examined. General cognitive abilities were re-tested and the MMSE score amounted to 30/30. Performance IQ was still impaired (PIQ = 65, -2.3 SD). The patient obtained pathological scores for the digit symbol substitution (-3 SD) and block design (-2 SD) while depressed scores were found for matrix reasoning (-0.7 SD), picture completion (-1.3 SD) and picture arrangement (-1.3 SD). The verbal IQ (VIQ = 98; -0.1 SD) and full scale IQ (FSIQ = 82; 1.2 SD) were within the normal range. The WMS-R index score for attention remained impaired (68, -2.13 SD) whereas mnestic capacities had increased significantly: the visual memory index increased to 111 (+ 0.73 SD), the verbal memory index to 101 (+ 0.07 SD) and the delayed recall index to 115 (+ 1 SD). A pathological result was again found for the WCST: the patient completed only 3 categories. Performance on the Trail Making Test slightly increased to average levels for the TMT-A (< pc. 30) and low average levels for the TMT-B (< pc. 21). A similar observation was made with regard to his performance on the Stroop Color Word Test: card 1: pc. <15, card 2: < pc. 10; card 3: < pc. 15. Performance on the Frontal Assessment Battery slightly improved to the normal range: 15/18. The patient obtained a defective score of 8 on the Middelheim Frontality Scale (MFS; cut-off = ≥5, De Deyn et al., 2005), which confirmed behavioral abnormalities related to apathy, blunting of affect as well as anosognosia. The clinical profile was consistent with a diagnosis of brainstem cognitive affective syndrome (Mariën & D'aes, 2015).

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Table 3.1. Overview of the neuropsychological investigations: case 1

GENERAL	May 2012 Score (±1SD or pc.)	June 2012 Score (±1SD or pc.)	October 2012 Score (±1SD or pc.)	Maximum or # trials[range]	Mean (SD)
Mini Mental State Examination	27 (-0.5)		30 (+0.8)	30	28 (1.9)
WAIS-III					
Wechsler Full Scale IQ (FSIQ)	**73 (-1.8)**		**82 (-1.2)**		**100 (15)**
Wechsler Verbal IQ (VIQ)	**83 (-1.1)**		**98 (-0.1)**		**100 (15)**
Information	11 (+0.3)		9 (-0.3)		10 (3)
Comprehension	4 (-2.0)		10 (0.0)		10 (3)
Digit Span	5 (-1.7)		11 (+0.3)		10 (3)
Arithmetic	7 (-1.0)		9 (-0.3)		10 (3)
Similarities	8 (-0.7)		10 (0.0)		10 (3)
Vocabulary	8 (-0.7)		12 (+0.7)		10 (3)
Wechsler Performance IQ (PIQ)	**61 (-2.6)**		**65 (-2.3)**		**100 (15)**
Digit Symbol Substitution	1 (-3.0)		1 (-3.0)		10 (3)
Picture Completion	5 (-1.7)		6 (-1.3)		10 (3)
Block Design	6 (-1.3)		4 (-2.0)		10 (3)
Picture Arrangement	2 (-2.7)		6 (-1.3)		10 (3)
Matrix Reasoning	5 (-1.7)		8 (-0.7)		10 (3)
MEMORY					
Wechsler Memory Scale-R	**66 (-2.3)**	**66 (-2.3)**	**68 (-2.1)**		**100 (15)**
Visual Memory	70 (-2.0)	80 (-1.3)	111 (+0.7)		100 (15)
Verbal Memory	80 (-1.3)	90 (-0.7)	101 (+0.1)		100 (15)
General Memory	73 (-1.8)		104 (+0.3)		100 (15)
Delayed Recall	61 (-2.6)	83 (-1.1)	115 (+1.0)		100 (15)
EXECUTIVE FUNCTIONS					
Wisconsin Card Sorting Test	**3 (-2.3)**		**3 (-2.3)**	**(64)128 trials**	**5.58 (1.1)**
Verbal Fluency					
Semantic Fluency	40 (-1.1)		49 (-0.5)		56.1 (15.1)
Stroop Color-Word Test					
Card I	126” (< pc. 1)		62” (pc. 15)		
Card II	163” (< pc. 1)		80” (pc. 10)		
Card III	188” (pc. 3)		128” (pc. 15)		
Trail Making Test					

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GENERAL	May 2012 Score (±1SD or pc.)	June 2012 Score (±1SD or pc.)	October 2012 Score (±1SD or pc.)	Maximum or # trials[range]	Mean (SD)
Part A	94" (< pc. 10)		41" (pc. 30)		
Part B	223" (< pc. 10)		99" (pc. 25)		
BEHAVIORAL					
Frontal Assessment Battery[a]	**13**		**15**	**18**	
Middelheim Frontality Scale[b]			**8**		
LANGUAGE					
Systematische Opsporing Schrijfproblemen	**8 (-4.8)**		**5 (-2.5)**		**1.7 (1.3)**
Comprehensive Aphasia Test					
Cognitive screening					
Semantic memory		10 (+0.3)		10 [7-10]	9.7 (0.9)
Word fluency		13			
Recognition memory		9 (-0.7)		10 [4-10]	9.6 (0.9)
Gesture object use		10 (-2.1)		12 [9-12]	11.5 (0.7)
Arithmetic		5 (-0.6)		6 [2-6]	5.5 (0.8)
Language comprehension					
Comprehension of spoken words		30 (+0.9)		30 [20-30]	28.5 (1.7)
Comprehension of written words		30 (+0.5)		30 [24-30]	29.5 (1)
Comprehension of spoken sentences		30 (-0.5)		32 [20-32]	30.8 (1.7)
Comprehension of written sentences		30 (-0.2)		32 [20-32]	30.4 (2.2)
Expressive language					
Repetition of digit strings		5 (-0.6)		7 [0-7]	5.6 (1)
Repetition of sentences		6 (+0.5)		6 [4-6]	5.8 (0.4)
Naming objects		46 (+1)		48 [25-48]	43.2 (4.5)
Naming actions		8 (-1.9)		10 [5-10]	9.5 (0.8)
Reading words		48 (+0.5)		48 [40-48]	47.3 (1.5)
Reading complex words		6 (+0.3)		6 [2-6]	5.9 (0.4)
Reading non-words		10 (+0.3)		10 [4-10]	9.6 (1.2)
Writing (copying)		31 (+0.3)		31 [5-31]	30.1 (3.1)

Abbreviations: FSIQ= Full Scale Intelligence Quotient, WMS-R= Wechsler Memory Scale-Rev, pc.= percentile, PIQ= Performance Intelligence Quotient, SD= Standard Deviation, VIQ= Verbal Intelligence Quotient

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Neurolinguistic assessments were carried out to further evaluate speech and language functions. The results revealed that the patient did not suffer from aphasia. Performance on the Psycholinguistic Assessments of Language Processing in Aphasia (PALPA Dutch version) (Bastiaanse et al., 1995) was within normal limits. The patient obtained normal results for the (Dutch) Comprehensive Aphasia Test (Dutch Version - CAT-NL) (Visch-Brink et al., 2015). The Test for Systematic Detection of Graphomotor Impairment (or 'Systematische opsporing schrijfmotorische problemen', *systematic detection of graphomotor problems*; SOS-test) (Van Waelvelde et al., 2012) revealed aberrant writing skills in May 2012 (score 8, -4.8 SD) and scores were indicative of an apraxic agraphia. The score was still depressed after retesting in October of the same year (score 5, -2.5 SD).

3.2.2. Case 2

Case 2 is a 72-year-old right-handed polyglot English man, who was admitted to hospital in October 2012 after an acute episode of balance problems, vertigo, and vomiting. On admission to the hospital he presented with a left-sided ataxia, dysarthria (slurred speech), and a tendency to fall over to the right side. Although the patient mastered six foreign languages [English (maternal language), French (learned in school at age 11), German (learned in school at age 13), Serbo-Croat (learned after crash course at age 24), Hebrew (Ivrit) (learned at an intensive course at age 28), and Dutch (moved to Belgium at age 35 and married a Flemish-speaking woman)] he could only speak and understand his native language, which was British English. Although he indicated that he had some very mild word-finding difficulties in English. He was completely unable to have a conversation in Dutch or any of the other non-native languages (= a differential polyglot aphasia). Medical history included arterial hypertension, coronary heart disease and a left occipital stroke 10 years before this admission. The patient had an educational level of 12 years (grammar school) and had worked as a (war) journalist in different countries. On admission, his speech was qualified as 'slurred' but over the course of days it became characterized by a slow rate with sudden accelerations, breathiness, altered word stress, and odd intonation patterns (terminal pitch rise). In terms of segmental changes, the diphthongs were backed and consonant articulation became unclear when speech rate increased. Although he severely struggled for everyday words and grammar, his command of Dutch gradually started to return two days post-stroke but did not reach premorbid proficiency levels. Minimal progress was made with regard to the other non-native languages. The patient's daughter reported that during the first days post-stroke her father had started speaking with an 'Estuary English' accent, typical of the region where the patient grew up. He had commuted between Australia and Europe for more than twelve years for his work. Consequently, he had adopted an Australian accent. In Dutch an accent change was found from pre-stroke standard Dutch accent to a regional accent (Antwerp).

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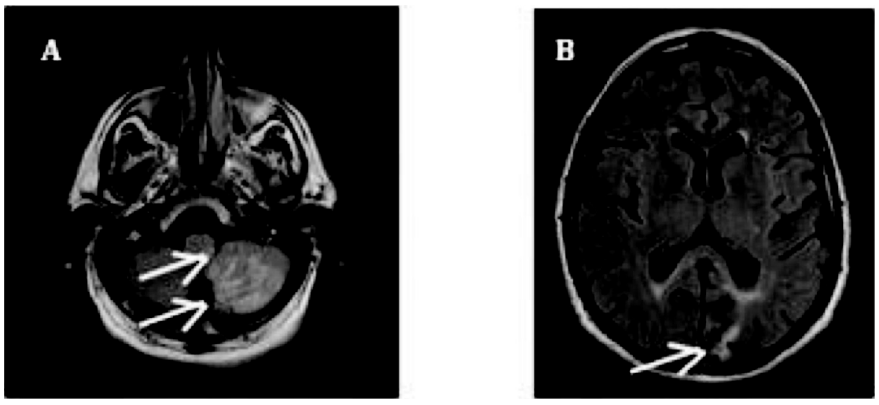
The T1- and T2-weighted axial FLAIR imaging of the brain showed inhomogeneous hyperintense areas in the left inferior cerebellar hemisphere and the postero-inferior left cerebellum, consistent with a recent infarction in the vascular territory of the left posterior inferior cerebellar artery (PICA). A hyperintense area was observed in the left posterior inferior brainstem (**Figure 3.3a**). In addition, an old vascular lesion was found in the left occipital lobe as well as small vascular lesions in the periventricular white matter (**Figure 3.3b**). MRA revealed atheromatosis of the distal left vertebral artery (no flow was detected) and the right distal vertebral artery and proximal basilar artery (irregular shape).

Figure 3.3. T2-weighted axial slice of the brain (a) at the level of the cerebellum, demonstrating (left) a hyperintense area in the left posterior inferior brainstem and postero-inferior portion of left cerebellar hemisphere and (b) demonstrating an old infarction in the calcarine region of the left occipital lobe.



In-depth neuropsychological investigations were performed (in English) one week after stroke (see **table 3.2**). A strong and consistent right hand preference was objectified by means of the Edinburgh Handedness Inventory (Oldfield et al., 1971) (laterality quotient = +100). General cognitive screening showed normal performance (MMSE: 27/30, SD: -0.6). The RBANS (Randolph, 1998) showed superior visuospatial/constructive skills (index 121, SD: +1.4) and a very superior immediate recall (index 136, SD: +2.4). Language (index 96, SD: -0.3), attention (index 94, SD: -0.4), and delayed recall (index 101, SD: +0.1) were within the normal range. Raven's Colored Progressive Matrices (Raven, 1976) revealed high average visuospatial problem solving skills (pc. 90). Frontal planning and problem solving were normal as well. The WCST showed normal cognitive flexibility and frontal problem solving (4 categories within 64 trials). The Stroop Color Word test (pc. 20) showed a low average ability to inhibit a competing and more automatic response.

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As evidenced by the CAT (Howard et al., 2004), the patient obtained maximum results when tested in his mother tongue. Visual confrontation naming on the BNT (Kaplan et al., 1983) was normal for English (57/60, SD: +0.3) but severely pathological for Dutch (BNT: 25/60, SD: -10.6): 13/35 errors were intrusions from English words (foreign words), whereas for 18/35 pictures no response was given. Semantic verbal fluency, assessed by means of a one-minute oral production of words belonging to a specific semantic category, was within the normal range (58 items, SD: +0.3).

A diagnosis of differential polyglot aphasia and FAS following a left cerebellar stroke was made. At the behavioral level no disturbances were noted. He remained positive and calm.

Table 3.2. Overview of the neuropsychological investigations: case 2

GENERAL	Score (± SD or pc.)	Maximum or #trials [range]	Mean (SD)
Folstein MMSE	**27 (-0.6)**	**30**	**28 (1.6)**
RBANS			
Visuospatial/constructive index	121 (+1.4)		100 (15)
Immediate recall index	136 (+2.4)		100 (15)
Language index	96 (-0.3)		100 (15)
Attention index	94 (-0.4)		100 (15)
Delayed recall index	101 (+0.1)		100 (15)
Raven's Colored Progressive Matrices	**29 (pc. 90)**	**36**	
EXECUTIVE FUNCTIONS			
Wisconsin Card Sorting Test	**4 cat**	**(64) 128 trials**	**4 cat**
Stroop Color Word Test			
card 1	45" (pc. 65)		48''
card 2	46" (pc. 95)		63''
card 3	128" (pc. 20)		99''
LANGUAGE			
Boston Naming Test-NL	**25 (-10.6)**	**60**	**54.9 (2.8)**
Boston Naming Test-EN	**57 (+0.3)**	**60**	**53 (7.3)**
Verbal Fluency	**58 (+0.3)**		**53.9 (13.2)**
Comprehensive Aphasia Test			
Cognitive screening			
Line bisection	-1 (+0.9)	-1 [-2;-1]	-0.2 (0.9)
Semantic memory	10 (+0.3)	10 [7-10]	9.7 (0.9)

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Recognition memory	10 (+0.4)	10 [4-10]	9.6 (0.9)
Gesture object use	10 (-2.14)	10 [9-12]	11.5 (0.7)
Arithmetic	6 (+0.6)	6 [2-6]	5.5 (0.8)
Language comprehension			
Comprehension of spoken words	30 (+0.9)	30 [20-30]	28.5 (1.7)
Comprehension of written words	30 (+0.5)	30 [24-30]	29.5 (1)
Comprehension of spoken sentences	32 (+0.7)	32 [20-32]	30.8 (1.7)
Comprehension of written sentences	32 (+0.7)	32 [20-32]	30.4 (2.2)
Comprehension of spoken paragraphs	4 (+0.7)	4 [0-4]	3.5 (0.7)
Expressive language			
Repetition of words	32 (+0.4)	32 [24-32]	31.4 (1.4)
Repetition of complex words	6 (+0.2)	6 [2-6]	5.9 (0.5)
Repetition of nonwords	10 (+0.6)	10 [0-10]	9 (1.8)
Repetition of digit strings	7 (+1.2)	7 [0-7]	5.6 (1.0)
Repetition of sentences	6 (+0.5)	6 [4-6]	5.8 (0.4)
Naming objects	48 (+1)	48 [25-48]	43.2 (4.5)
Naming actions	10 (+0.6)	10 [5-10]	9.5 (0.8)
Reading words	48 (+0.5)	48 [40-48]	47.3 (1.5)
Reading complex words	6 (+0.3)	6 [2-6]	5.9 (0.4)
Reading function words	6 (+0)	6 [4-6]	6 (0.2)
Reading non-words	10 (+0.3)	10 [4-10]	9.6 (1.2)
Writing (copying)	31 (+0.3)	31 [5-31]	30.1 (3.1)
Written picture names	23 (+0.7)	23 [8-23]	21.6 (2)

Abbreviations: MMSE= Mini Mental State Examination, pc.= percentile, RBANS= Repeatable Battery for the Assessment of Neuropsychological Status, SD= Standard Deviation

3.3. SURVEY OF THE LITERATURE: FAS AFTER POSTERIOR FOSSA LESIONS

Cole (1971) was the first to describe two patients with FAS after posterior fossa lesions (Appendix B1, case 1 and 2). The first patient was a 29-year-old polyglot man, who lived in Ohio and spoke American English and Yiddish. He developed FAS *presumably* in the context of cerebellar degeneration due to anoxia. The neurological symptoms consisted of dysphagia, ataxia of arms and legs, spastic-ataxic gait, and a bilateral Babinski sign. He suddenly developed an accent, which was perceived as Italian. The second

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patient was a 58-year-old right-handed, polyglot (English/Hungarian) woman, who suffered an inferior pontine infarction. Post-stroke she developed an Eastern European accent. Initial symptoms included vertigo, diplopia, horizontal gaze palsy, speech difficulties (a FAS and a dysarthria), and ataxia of the arms and legs. Before the stroke, friends and family considered her accent to be American English, typical of Ohio, where she was a long-time resident. Cole (1971) questioned whether there is a relationship between FAS and dysarthria and whether the alterations of speech prosody and rhythm are related to posterior fossa lesions. Robert Joynt commented on these unexpected findings as follows: "...it doesn't appear unusual that any of the brain mechanisms which are used to integrate motor acts may alter force, prosody, and rhythm of speech. It is well that Dr. Cole has pointed this out with posterior fossa lesions" (p.153). However, he ironically added: "(...) I am enough of a romanticist to think that these subtle inflections and nuances of speech must ultimately stem from the cerebral cortex. For example, if Juliet had said in a monotone, "Romeo, Romeo, wherefore art thou, Romeo?" I doubt if Romeo would have done himself in for her. But then, I don't think Shakespeare wrote for the cerebellum." (p.153). In the discussion it was concluded that distinct forms of dysarthria-like syndromes exist: those due to posterior fossa lesions and those due to frontal lesions.

Forty years later, Dankovičová & Hunt (2011) (Appendix B1, case 3) described a 56-year-old man with a stroke affecting the anterior margin of the mid pons immediately left to the midline. The lesion was discovered on MRI 16 months after the patient suffered the stroke. The patient suffered from persistent speech difficulties and a change of accent (identified as Italian or Greek in a perceptual accent rating experiment). The patient's original accent was typical of his Essex hometown. He was raised in London. His father was an Englishman and his mother was of Greek descent. However, she declared that she had always spoken English with her son. In the light of the patient's speech characteristics, the authors hypothesized that FAS after a pontine lesion may be related to dysarthria (of the ataxic or bulbar/flaccid-type).

In 2013, Tran & Mills (Appendix B1, case 4) described a 60-year-old African-American woman, who presented at the emergency department with an acute change of accent that appeared to her as Jamaican. She equally experienced gait disturbances, heaviness in both legs and had a severe headache. A CT-scan of the brain on admission was normal but an MRI-scan revealed an infarction in the left pons. Magnetic Resonance Angiography (MRA) confirmed this finding.

In one case, a lesion in the posterior fossa was argued to be – counter intuitively – implicated in the accent *remission*. In 2009, Cohen et al. (Appendix B1, case 5) described a 58-year-old English-speaking woman who developed FAS after a left frontoparietal infarction. However, she suddenly *lost* the accent after incurring a right cerebellar hemorrhage. Cohen et al. explained this phenomenon stating that the infarction may have caused a disruption of the "competitive interactions between the two cerebellar hemispheres" (p. 566) and may have led to functional facilitation in the left cerebellar

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hemisphere in the presence of "maladaptive activation" (p. 566) in the right cerebellar hemisphere.

The case described by Mariën et al. (2013) (Appendix B1, case 6) suffered a cerebellar hemorrhage in the right cerebellar hemisphere, demonstrated on a CT and an MRI. The stroke receded without intervention but left the patient with a posterior fossa syndrome and cerebellar cognitive affective syndrome (CCAS). Initially the patient was mute (akinetic), although later he developed FAS and an ataxic dysarthria. Due to the important cognitive and linguistic impairments as well as the personality changes the researchers decided to perform a SPECT scan. The important crossed cerebro-cerebellar connections were affected and a severe hypoperfusion was demonstrated in the bilateral prefrontal areas, which could account for the executive disturbances and behavioral changes, and was reconcilable with the linguistic profile.

Three patients demonstrated FAS after a disruption of the functional connection between the perisylvian speech areas and the infratentorial regions (i.e. the cerebellum) (Hwang et al., 2001; Verhoeven & Mariën, 2002; Mariën & Verhoeven, 2007, case 2). Hwang et al. (2001) (Appendix B2, case 1) described a 40-year-old, right-handed, Mandarin-Chinese speaking Taiwanese woman, who was admitted to hospital after a sudden change of her native accent to (American) English as (an isolated symptom). Initial physical and neurological examinations, as well as blood tests were normal. The patient displayed focal hyperintensities at the level of the bilateral internal capsules (genua and posterior limbs) on a T2-weighted MRI, but these were considered clinically insignificant. The accent receded three days after her hospitalization, but the hyperintensities were still visible on a repeat MRI two years later. In contrast, SPECT performed 1 day after admission revealed a hypoperfusion at the level of the left lateral temporal region and right cerebellum. The authors hypothesized that these functional deficits were due to a transient ischemic attack in the left temporal area. Repeat SPECT two years later showed a complete resolution. Hence, the authors argued that the hypoperfusion may have played a significant role in the accent change.

The case described by Verhoeven & Mariën (2002), also discussed in Mariën & Verhoeven (2007) (Appendix B2) is the first of two stroke patients in whom the remission of FAS symptoms was directly related to normalization of the hypoperfusion in the right cerebellum after focal left frontoparietal damage (diaschisis). The patient was a 53-year-old, right-handed, Dutch-speaking Belgian woman who was admitted to hospital because of an acute motor speech impairment rapidly evolving to mutism associated with a right-sided loss of strength. In addition to mutism, the neurological examination revealed a right hemiparesis and central facial nerve palsy. The CT scan demonstrated an infarction in the left frontoparietal region. Two weeks later, an MRI confirmed ischemic damage in the left middle cerebral artery territory, comprising the inferior frontal gyrus, precentral gyrus, anterior insular cortex, postcentral gyrus and supramarginal gyrus. 99mTc ECD SPECT at 33 days post-stroke revealed a hypoperfu-

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sion in the left frontal motor and parietal cortex, the thalamus and striatum of the left hemisphere as well as in the right cerebellum (no standard deviations provided). Repeat SPECT three years after the stroke showed that the perfusion deficits at the supratentorial level had only marginally improved, but that the perfusion in the right cerebellum had normalized. Parallel to the observed improvement of the perfusion changes at the cerebellar level, a resolution of FAS (French or Russian-like accent) was attested. The second patient was a 61-year-old right-handed Dutch-speaking Belgian man who suffered a basal ganglia hemorrhage (Appendix B2, case 3). This patient acutely developed a global aphasia, which evolved to a conduction aphasia after approximately one week. After this transition of aphasic symptoms, the patient's accent was perceived as 'North-African'. Neurological examinations disclosed a right hemiplegia and right hemianopia. A repeat MRI of the brain three weeks later confirmed that the lesion comprised the left putamen, genu and posterior limb of the internal capsule extending to the insula, temporal and parietal lobe. A quantified 99mTc ECD SPECT one month post-stroke revealed a severe hypoperfusion (more than 2 SD) at the cortical and subcortical level (including the left thalamus, left lentiform nucleus, left medial, lateral temporal region, left motor cortex and right cerebellum). Repeated SPECT completed at six months post-stroke demonstrated a complete resolution of the hypoperfusion at the infratentorial level, while perfusion deficits at the supratentorial level were only slightly enhanced. Around the same time (i.e. 6 months post-stroke) an improvement in both the FAS and aphasic symptoms was observed. The congruent remission of the cerebellar hypoperfusion and the linguistic deficits left the authors to hypothesize on the role of the cerebellum in motor speech (planning) disorders, and hence also FAS.

3.4. DISCUSSION

Two new cases of FAS after a posterior fossa lesion are documented. The first case developed a FAS and a paralytic dysarthria after an ischemic stroke in the right pons, disrupting brainstem nuclei and interrupting the crossed ponto-cerebellar pathway. The combination of linguistic, cognitive and affective changes allowed for a diagnosis of "brainstem cognitive affective syndrome" which closely relates to Schmahmann's syndrome (Manto & Mariën, 2015). From an anatomoclinical point of view the clinical symptoms were consistent with the SPECT findings, demonstrating a hypoperfusion affecting the frontal and prefrontal areas bilaterally as well as in the left cerebellum (Mariën & D'aes, 2015). The second case developed a differential polyglot aphasia and FAS after an infarction in the vascular territory of left PICA. He did not demonstrate any affective or behavioral disturbances, nor any cognitive impairments. At the linguistic level, however, Dutch as well as all the other non-native languages were severely disturbed while his mother tongue (British English) remained clinically unaffected (differential polyglot aphasia). Post-stroke FAS reflected a reversion to a previously learned accent (Seliger et al., 1992; Roth et al., 1997). Both patients' speech characteristics may

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equally be associated with apraxia of speech (AoS) (slow speech rate, increase in errors when speech rate is increased, attainment of vowels more than consonants, (irregular) initiation difficulty) or ataxic dysarthria (slow speech rate altered with sudden fast speech rate, disrupted articulation of vowels and consonants, slurring) (Duffy, 2013), but do not allow for a clear differentiation. Ataxic dysarthria is the most typical motor speech disorder associated with cerebellar disorders and is mainly characterized by a distorted articulation and prosody (Spencer & Slocomb, 2007). Although ataxic dysarthria is traditionally considered a disorder of motor execution, recent studies have attributed typical characteristics of ataxic dysarthria to a disruption of motor speech programming/planning instead of to a dysfunctional execution of speech (Mariën & Verhoeven, 2007; Spencer & Slocomb, 2007). Since ataxic dysarthria shares some evident semiological characteristics with AoS, a typical speech planning and programming disorder, some authors also hypothesized that ataxic dysarthria may be subserved by a similar pathophysiological mechanism (Mariën et al., 2006). However, only a handful of cases of AoS have been reported with possible cerebellar involvement. Apart from the FAS case of Verhoeven & Mariën (2002) (Appendix B2, case 2), one other case of AoS has been reported in which the right cerebellum may have played a role. Mariën et al. (2001) reported a patient who presented with agraphia and severe AoS after a left cerebral hemisphere infarction with lesions affecting the anterior insular cortex and the opercular part of the left inferior frontal gyrus. The SPECT scan not only showed a hypoperfusion in the insular region, but also in the right cerebellar hemisphere. After one year, AoS completely receded. This was reflected by a reperfusion of the right cerebellar hemisphere.

The case described by Mariën et al. (2013), however, demonstrated ataxic dysarthria, in combination with FAS (and CCAS) after a right cerebellar stroke. The newly reported patients demonstrated FAS in association with a *left* cerebellar hypoperfusion (case 1) and a *left* cerebellar infarction (case 2). It can be hypothesized that the disruption of the cerebro-cerebellar network – due to a pontine (case 1) and left cerebellar lesion (case 2) – is responsible for the planning and organization deficits at both the linguistic and non-linguistic level. The first patient presented a significant hypoperfusion of the left inferior medial frontal regions as a result of cerebello-cerebral diaschisis reflecting the loss of excitatory impulses from the cerebellum to the supratentorial areas subserving the planning and organization of motor and non-motor linguistic and non-linguistic processes, including articulation. For the second patient, no SPECT data were available. Speech production, and more particularly speaking with a monotonous voice, has not only been associated with right, but also left cerebellar hemisphere activations in overt and covert speech tasks (Dogil et al., 2002). The differential resolution of the aphasia selectively affecting the non-native languages in this patient, in combination with the attested left cerebellar damage, could be related to the theories of the more bilaterally distributed speech and language networks possibly also those mediating prosody in particular in bilinguals and polyglots. Especially more right hemisphere structures are recruited in polyglots when speaking non-native languages. These regions

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are structurally connected with the contralateral left cerebellar hemisphere, from which this patient possibly received less excitatory impulses (Meschyan & Hernandez, 2006; see also Abutalebi & Green, 2016).

From a pathophysiological perspective, FAS has been related to an impaired position of vocal tract or increased tension (Kurowski et al., 1996; Ingram et al., 1992). If the increased tension would be one of the primary instigators of FAS-like speech, then this could be an argument in favor of FAS as an executive disturbance: the increased muscle tone can be an upper motor neuron sign and characterizes patients with (spastic/pseudobulbar) dysarthria. This would constitute an "organic explanation" for FAS. However, current patients both presented with dysarthria types associated with muscle tone weakness: paralytic dysarthria and ataxic dysarthria. This explanation does not seem applicable for current patients. From a neurocomputational viewpoint, Tomasino et al. (2013) argued that the deficits of FAS patients are due to deficient sensory feedback (in the case of primarily subcortical damage) or feedforward (in the case of cortical and perilesional subcortical damage) control mechanisms. They explain deficits in the context of the DIVA-model (i.e. Directions into Velocity of Articulators) (Golfinopolous et al., 2010; Tourville & Guenther, 2011). The model describes speech production as relying on two subsystems: a feedforward control system and a feedback control system. Applied to speech development, the model can be explained as follows: the phonemes that were acquired at early age are stored in speech sound maps in the inferior frontal gyrus and are directly linked to articulator position maps stored in the ventral premotor cortex; together they form the feedforward control loop. Via the feedforward control, one is aware of the phonological results of set of motor programs (predictive component) that are transferred to the articulator position and velocity maps. When the initiation maps in the (bilateral) supplementary motor area become active, these commands are released. In these commands the position of the different articulatory variables (jaw height, tongue height, tongue shape etc.) are encoded, which are translated into movements of the vocal tract. The speech sound maps define the auditory and somatosensory target maps in the feedback control system. When speech is articulated, the somatosensory and auditory-perceptual awareness, mediated by subcortical nuclei, activates the feedback control loop, which allows for an update of the feedforward loop and will try to correct articulatory encoding to achieve these targets correctly next time. If the deficits in FAS are explicable, even partly, in the context of the DIVA-model then a role for the cerebellum – affected in both patients is not inconceivable. The cerebellum plays an important role in "the learning and maintenance of feedforward motor commands" (Tourville & Guenther, 2011, p. 11]. Since it stands in close connection to the motor areas on the one hand, and on the other hand receives input from the somatosensory cortex and auditory areas (Tourville & Guenther, 2011), it is attributed a cardinal role in feedforward motor control via supposed feedback motor learning in response to a miscalibration of intended movement or execution targets (Schmahmann & Pandya, 1997). These feedforward projections are hypothesized also include the basal

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ganglia (axons in the dentate nucleus in the cerebellum projects to the contralateral (motor) cortex via the thalamus). This is reconcilable with the theories of Schiff et al. (1983), Reeves et al. (2007) and Blumstein & Kurowski (2006) who claim that FAS is due to a disruption of the cortico-striatopallidal-thalamic pathways. Thus the rhythmic disturbance can be consistent with a defect in the predictions (feedforward control) exerted by the cerebellar circuitry. This could then also serve as an explanation for the previously hypothesized semiological association between FAS and AoS, and by inference FAS, AoS, and ataxic dysarthria. The hypothesis of errors in predictions leading to compensatory substitution mechanisms by the unaffected cerebral areas (including the cerebellum itself) could be put forward. From a cognitive perspective, FAS has been viewed as the result of an impairment in distinguishing between a direct and indirect speech encoding route (Whiteside & Varley, 1998). The results of the SPECT scan for the first patient demonstrated disturbed physiological communication between the cerebellum and frontal regions, which are responsible for speech planning and programming. For the second patient, the left cerebellar hemisphere was damaged. Here a maladaptive compensation under the form of an aberrant recovery process (Manto et al., 1998) or an overcompensation mechanism, as reported when the inhibitory signals from the cerebellum are affected, could also be considered (Baier et al., 2015). A maladaptive compensation from the part of the contralateral cerebellar hemisphere, exerting distance effects on the frontal cerebral areas (including for instance the motor cortex), can be hypothesized. The precise roles of (a) the olivo-cerebellar pathway, an inflow tract to the cerebellum with a major role in pace-making activity, and (b) the projections from the cerebellar cortex to cerebellar nuclei remain undetermined. However, these explanations are reminiscent of the case described by Cohen et al. (2009) who described a 58-year-old woman who developed FAS after an MCA stroke affecting the left frontoparietal lobe. The authors dwell on how the sudden remission after a second cerebellar stroke in the right inferior posterior lobe can be explained. Cerebellar-parietal connections have been argued to underpin phonological storage, although its exact role remains unclear (Macher et al., 2014; Ziegler, 2016).

In conclusion, it seems that a disruption of the cerebro-cerebellar circuitry following a right pontine lesion and left cerebellar lesion in the posterior lobe, exerting remote functional effects in the cortical areas subserving planning and organization, may play a foremost important role in the pathophysiology of FAS following posterior fossa lesions. Several hypotheses have been formulated as to how this damage may be related to the onset of FAS. These explanations were not mutually exclusive. It was argued that FAS can be the consequence of disturbed feedforward and/or feedback speech control mechanisms. The cerebellum has been hypothesized to play an important role in feedforward speech control via projections to the motor cortex. This was relatable to earlier theories on the pathophysiology of FAS. Latter theories can explain the FAS in the first case: the crossed ponto-cerebellar pathway (inflow tract to the cerebellum) was damaged, which had clear repercussions on the (pre)frontal areas as was demonstrated by a SPECT scan. On the other hand, the possibility of maladaptive compensation from the

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part of, possibly, the contralateral cerebellum could be related to the FAS characteristics in case 2. However, this patient was a polyglot. Hence, the impact of the left cerebellar hemisphere lesion on the speech and language in this patient should also be considered in the context of the more bilaterally distributed speech network in polyglots.

In future studies on FAS, DTI and/or fMRI experiments may be a relevant addition with a view to investigate (possibly disrupted) (bi-hemispheric) network activity, as well as fiber tracts. This will allow researchers to hypothesize on FAS prognosis and possibly also on whether the semiological resemblance with (ataxic) dysarthria or AoS, is grounded on a shared neural and/or neurophysiological substrate.

3.5. ACKNOWLEDGMENTS

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PART II: DEVELOPMENTAL FAS

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CHAPTER 4

Developmental Foreign Accent Syndrome: Report of a New Case[10]

[10] The study reported in this chapter was published in Frontiers in Human Neuroscience: Keulen, S., Mariën, P., Wackenier, P., Jonkers, R., Bastiaanse, R., & Verhoeven, J. (2016) Developmental foreign accent syndrome: report of a new case. Frontiers in Human Neuroscience, 10, 1-9.

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ABSTRACT

This paper presents the case of a 17-year-old right-handed Belgian boy with developmental FAS and comorbid developmental apraxia of speech (DAS). Extensive neuropsychological and neurolinguistic investigations demonstrated a normal IQ but impaired planning (visuo-constructional dyspraxia). A Tc-99m-ECD SPECT revealed a significant hypoperfusion in the prefrontal and medial frontal regions, as well as in the lateral temporal regions. Hypoperfusion in the right cerebellum almost reached significance. It is hypothesized that these clinical findings support the view that FAS and DAS are related phenomena following impairment of the cerebro-cerebellar network.

4.1. INTRODUCTION

Foreign accent syndrome (FAS) is a relatively rare motor speech disorder in which segmental and prosodic speech alterations cause patients to be perceived as non-native speakers of their mother tongue (Blumstein et al., 1987; Lippert-Gruener et al., 2005; Tran & Mills, 2013; Pyun et al., 2013; Ingram, 1992). In some cases, there is a reversion to a previously acquired language variety (Seliger, 1992; Kwon & Kim, 2006). In 2010, Verhoeven & Mariën provided a taxonomical classification of this speech disorder and defined three main types of FAS: a neurogenic, psychogenic and mixed type (Verhoeven & Mariën, 2010a). Neurogenic FAS is further subdivided into an acquired and a developmental[11] variant. The current article focuses on developmental FAS, which is one of the rarest etiological subtypes of FAS. To the best of our knowledge only two case studies have been published between 1907 and 2014 (Mariën et al., 2009). The first case was a 29-year-old female native speaker of Belgian Dutch who was diagnosed with FAS and developmental apraxia of speech (DAS). The second patient was a 7-year-old boy, who presented with FAS in the context of specific language impairment (SLI) of the phonological-syntactic type (Mariën et al., 2009).

Although the number of documented developmental FAS cases has remained low, accent change has been (anecdotally) reported in relation to neurodevelopmental disorders, especially autism of the Asperger-type (Ghazziudin, 2005; Tantam 2012; Garnett & Atwood, 1997). However, in these reports, the neurobiological relationship between the speech characteristics and the developmental disorder was not addressed in detail. Hence, it is possible that FAS is much more common in a population with developmental disorders than current statistics indicate. This article presents a new case of developmental FAS in combination with DAS: a neurologically based speech disorder that affects the planning/programming of phonemes and articulatory sequences as langu-

[11] As the focus of the current article is the developmental subtype of foreign accent syndrome, the interested reader is referred to Verhoeven and Mariën (2010a) for a comprehensive discussion of the FAS taxonomy.

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age develops, in the absence of neuromuscular impairment (McNeill & Kent, 1990; Crary, 1984; Smith et al. 1994). The patient is a 17-year-old right-handed native speaker of Belgian Dutch (Verhoeven, 2005) who presented with articulatory problems and an accent, which was perceived as French or 'Mediterranean' by family, medical staff and acquaintances. A neurological and neuropsychological assessment was carried out and both an MRI and a SPECT were performed. Furthermore, the patient's speech was analyzed phonetically. Since this occurrence of FAS is linked to a programming disorder, the hypothesis of FAS as a possible subtype of apraxia of speech will be addressed in detail.

4.2. BACKGROUND

The assessment presented in this article was carried out following the principles of the standard clinical neurolinguistic work-up of patients with speech- and/or language disorders at ZNA Middelheim hospital in Antwerp (Belgium). The patient's parents provided written informed consent to report the patient's medical data.

A 17-year-old, right-handed, native speaker of Belgian Dutch consulted the department of Clinical Neurolinguistics of ZNA Middelheim Hospital because of persisting articulation difficulties resulting in accented speech. The patient indicated that listeners identified him as a non-native speaker of Dutch with a French or 'Mediterranean' accent. He was born at term after normal gestation and labor, and there had been no perinatal or postnatal problems. Medical history was unremarkable. According to 'WHO child growth standards' acquisition of gross motor milestones was normal. He could sit without support at 5.5 months (mean = 6.0; SD = 1.1), stand with assistance at 7 months (mean = 7.6; SD = 1.4) and walk independently at the age of 11 months (mean = 12; SD = 1.8 months). He was able to independently ride a bicycle without support at the age of 4.0 years. By the age of 4-5 years he had developed a clear right-hand preference.

Except for a deviant development of articulation skills, developmental milestones were normal, including non-motor speech and language ability. The patient did not present with any pervasive developmental disorder and no family history of developmental disorders or learning disabilities was reported. There were no clinical indications for a psychiatric disorder. The parents and close relatives stated that the patient was in perfect mental health. The patient was not under any medication at the time of examination. Speech therapy was started at the age of 5 years and discontinued at the age of 10 because of a lack of therapeutic progress. The parents were monolingual speakers of Dutch. The patient had successfully finished primary school and obtained above average results in the 3rd grade of secondary school. Neurological investigations, including EEG recordings, were normal. MRI of the brain revealed no lesions at the supra- and infratentorial level. There was no brain atrophy.

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A quantified Tc-99m-ECD SPECT study was carried out. 740 MBq (20 mCi) Tc-99m-ECD was administered to the patient by means of a previously fixed butterfly needle while he was sitting in a quiet dim room, eyes open and ears unplugged. Acquisition was started 40 min after injection using a three-headed rotating gamma camera system (Triad 88; Trionix Research Laboratory, Twinsburg, Ohio, USA) equipped with lead super-fine fanbeam collimators with a system resolution of 7.3 mm FWHM (rotating radius 13 cm). Projection data were accumulated in a 128 x 64 matrix, pixel size 3.56 mm, 15 seconds per angle, 120 angles for each detector (3° steps, 360° rotation). Projection images were rebinned to parallel data, smoothed and reconstructed in a 64 x 64 matrix, using a Butterworth filter with a high cut frequency of 0.7 cycles/cm and a roll-off of 5. No attenuation or scatter correction was performed. Trans-axial images with a pixel size of 3.56 mm were anatomically standardized using SPM and compared to a standard normal and SD image obtained from ECD perfusion studies in a group of 15 normally educated healthy adults consisting of 8 men and 7 women with an age ranging from 45 to 70 years. This normal image was created by co-registration of each normal study to the SPECT template image of SPM using the 'normalize' function in SPM. At the same time, the global brain uptake of each study was normalized. On the mean image, 31 ROI's were drawn and a 31 ROI template was created. Using the normalized studies and the 31 ROI template, the mean normal uptake and SD value (=1 Z-score) in each ROI was defined. Patient data were normalized using SPM in the same way and the perfusion uptake in each ROI was calculated. From this uptake, the mean uptake and SD value of the normal database, the Z-score for each region can be calculated. A regional Z-score of >2.0 is considered significant. SPECT findings are illustrated in **figure 4.1**:

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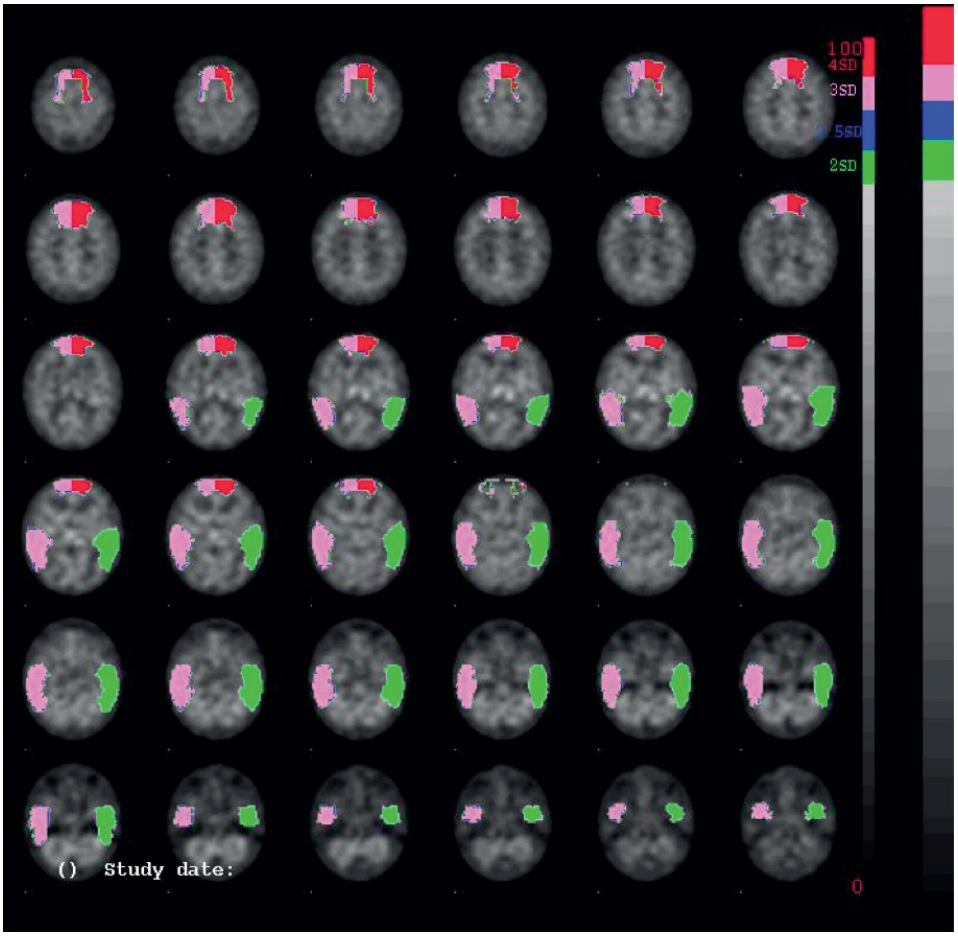
Figure 4.1. SPECT-findings demonstrating a significant decrease of perfusion bilaterally in the prefrontal and medial frontal regions, as well as in the lateral temporal regions.



A significant bilateral hypoperfusion distributed in the medial prefrontal regions (right: -3.48 SD; left: -4.97 SD) and in both lateral temporal regions (right: -3.17 SD; left: -2.17 SD) was found. Decreased perfusion in the left inferior medial frontal region (-1.65 SD), the right inferior lateral frontal region (-1.62 SD) and the right cerebellar hemisphere (-1.52 SD) nearly reached significance.

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4.3. METHODS

4.3.1. Neuropsychological Investigations

In-depth neuropsychological assessment consisted of a range of formal tests including the Wechsler Adult Intelligence Scale, 4th Ed., Dutch version (WAIS-IV-NL) (WAIS-IV: Wechsler, 2008; WAIS-IV-NL: Kooij & Dek, 2012), the Bourdon-Vos Test (Vos, 1998), the Wisconsin Card Sorting Test (WCST) (Heaton et al., 1993), the Stroop Color-Word Test (Stroop, 1935; Golden, 1978), the Trail Making Test (TMT) (Reitan, 1958), the Rey-Osterrieth figure (Rey, 1941; Osterrieth, 1944), the praxis subtests of the Hierarchic Dementia Scale (HDS) (Cole & Dastoor, 1987), the Beery Developmental Test of Visual-Motor Integration, 5th Ed. (Beery & Beery, 2004) and the Test of Visual-Perceptual Skills, third edition (TVPS-3) (Martin, 2006). Neurolinguistic assessment consisted of the Boston Naming Test (Kaplan et al., 1983; Belgian norms (Dutch): Mariën et al., 1998), the Clinical Evaluation of Language Fundamentals (Dutch version) (Semel et al., 2003) and the Dudal Spelling Tests (Dudal 1998, 2004). Test results are summarized in **Table 4.1.**

Table 4.1. Overview of the neuropsychological test results

TEST	Scaled score (raw score)	Percentile	Mean	SD	*Z-score*
INTELLIGENCE (WAIS IV)					
Wechsler Full Scale IQ (FSIQ)	119		100	15	+1.27
Wechsler Verbal Comprehension Scale	122		100	15	+1.47
Similarities	13		10	3	+1
Vocabulary	15		10	3	+1.67
Information	14		10	3	+1.33
Wechsler Perceptual Reasoning Scale	112		100	15	+0.8
Block Design	10		10	3	0
Matrix Reasoning	14		10	3	+1.33
Visual Puzzles	12		10	3	+0.67
Wechsler Working Memory Scale	117		100	15	+1.33
Digit Span	12		10	3	+0.67
Arithmetic	14		10	3	+1.33
Wechsler Processing Speed Scale	103		100	15	+0.2
Symbol Search	11		10	3	+0.33
Coding	10		10	3	0
MEMORY					
WMS-R Visual Memory Index	120		100	15	+1.33
Figure Memory	(8/10)				
Visual Paired Associates I	(18/18)				
Visual Reproduction I	(39/41)	92			
WMS-R Verbal Memory Index	126		100	15	+1.73
Logical Memory I	(42/50)	98			
Verbal Paired Associates I	(22/24)				

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TEST	Scaled score (raw score)	Percentile	Mean	SD	*Z-score*
WMS-R General Memory Index	131		100	15	+2.06
WMS-R Delayed Recall Index	>138		100	15	>+2.53
Logical Memory II	(40/50)	97			
Visual Paired Associates II	(6/6)				
Verbal Paired Associates II	(8/8)				
.Visual Reproduction II	(39/41)	95			
ATTENTION					
Bourdon-Vos Test					
Speed	(9.87")	50	50		0
Accuracy	(2)	75	1.40	0.89	0.67
EXECUTIVE FUNCTIONS					
Wisconsin Card Sorting Test					
Nr of categories realized	(1)				
Nr of trials	(128)				
Stroop Color-Word Test					
Card I	(45")	50	45		0
Card II	(55")	50	55		0
Card III	(96")	30	95.70	0.58	-0.52
Trail Making Test					
Part A	(21")	>90			
Part B	(43")	>90			
LANGUAGE					
Boston Naming Test	(55/60)		47.89	4.31	+1.65
EMT-B	9		10	3	-0.33
EMT-B item 50	9		10	3	-0.33
Dudal spelling					
Words	(31/40)	55			+0.13
Sentences	(33/40)	80			+0.84
Total	(64/80)	70			+0.52
CELF-IV-NL					
Recalling Sentences	11	63	10	3	+0.33
Formulated Sentences	14	91	10	3	+1.33
Word Definitions	13	84	10	3	+1
Word Classes Receptive	16	98	10	3	+2
Word Classes Expressive	13	84	10	3	+1
Word Classes Total	15	95	10	3	+1.67
Understanding Spoken Paragraphs	14	91	10	3	+1.33
Sentence Assembly	14	91	10	3	+1.33
Semantic Relationships	13	84	10	3	+1
Core Language Index	121		100	15	+1.4
Receptive Language Index	129		100	15	+1.93
Expressive Language Index	118		100	15	+1.2
Language Content Index	125		100	15	+1.67
Language Structure Index	122		100	15	+1.47

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TEST	Scaled score (raw score)	Percentile	Mean	SD	*Z-score*
PRAXIS					
Rey Complex Figure	(28/36)		35	3	-2.33
HDS Ideatonal: It. 5	(10/10)		9.79	0.17	+1.24
HDS Ideomotor: It. 3	(10/10)		9.94	0.23	+0.26
VISUAL COGNITION					
Beery Visual-Motor Integration	78		100	15	-1.47
Beery Visual Perception	94		100	15	- 0.4
Beery Motor Coordination	73		100	15	-1.8

General cognitive skills as measured by the WAIS-IV showed a high average full scale IQ level (FSIQ = 119) and average to above average results for each of the subscales. Problems primarily concerned abstract concept formation: shifting and maintaining goal-oriented cognitive strategies in response to changing environmental contingencies was abnormal as the patient only succeeded to complete 1 category within 128 trials (WCST). The planning and construction of a complex geometrical form (Rey-Osterrieth figure) was abnormal. On the Beery Developmental Test of Visual-Motor Integration the patient obtained borderline results for visual-motor integration skills (-1.47 SD) and for visual-motor coordination (-1.8 SD). Visual perception was normal. Articulation and prosody in conversational and spontaneous speech were clearly abnormal. The patient produced several substitution errors as well as omissions and additions during spontaneous conversation. Oral-verbal diadochokinesis was within normal limits, whereas rapid repetition of polysyllabic words was hesitant. Visual confrontation naming (BNT) and semantic verbal fluency were normal as well. Indices on CELF-IV-NL (Semel et al., 2003) were all above average. No grammatical errors, and lexical retrieval difficulties were observed. Spelling of words and sentences (Dudal spelling) was normal. The isolated motor speech impairments consisted of substitution errors for consonants (affecting place and manner of articulation: e.g. 'groepje*n*' instead of 'groepje*s*': little groups, the use of a uvular trill instead of an alveolar trill) and vowels (affecting vowel distinctiveness), difficulties initiating words ('ra.. ra.. ra… geraak': get somewhere) and omissions of consonants ('geraa' instead of 'geraak', 'pagia' instead of 'pagina': page). These errors are consistent with a diagnosis of DAS (see also *'phonetic analysis'* below).

4.3.2. Phonetic analysis

A perceptual error analysis of a 1:36 min spontaneous speech sample consisting of 397 words was carried out. This was supplemented by an acoustic analysis of some key aspects of speech. As far as consonant production is concerned, occasional voicing errors were observed (*stravde* for *strafte:* past tense of 'punished'). It was furthermore striking that the speaker used a uvular trill instead of the alveolar trill: although both are acceptable realizations of the trill in Dutch, the alveolar trill is the more common variant in

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the Brabantine geographical region of origin of this speaker. It is precisely the usage of a uvular trill that is typical of French non-native speakers of Dutch.

With respect to vowel articulation, various distortions were observed. In order to quantify these deviations, the formant frequencies of the 358 peripheral vowels in the speech sample were measured by means of the signal processing software PRAAT (Boersma & Weeninck, 2015). The instances of schwa were not analyzed. The mean formant values of the FAS vowels are illustrated in **figure 4.2**. They have been correlated to the vowel formants of a group of 5 male native control speakers of Dutch from the same geographical region as the FAS speaker. The formant values of the control speakers were obtained in a data collection independent of this investigation, which is described in more detail in Adank et al. (2004).

Figure 4.2. Mean formant values of F1 and F2 (in Hz) of the Dutch vowels in the FAS speaker (filled circles) and the control group (unfilled circles). The lines connect the vowel realizations of the FAS speaker and the control group.

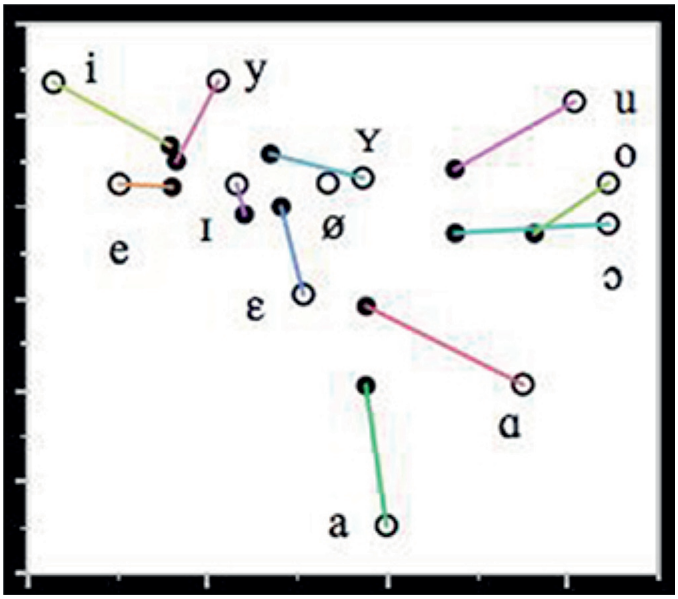

Figure 4.2 shows that with respect to vowel production: (1) there is a significant degree of vowel reduction and (2) a substantial erosion of vowel distinctiveness particularly in the front vowels. The observed vowel reduction, i.e. the more central realization of the vowels with respect to the control vowels, can be accounted for by the fact that the vowels in the FAS speaker and the control group have been recorded in different communicative settings. The vowels of the control group were recorded in a structured

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reading task in which the vowels were positioned in a prominent utterance position in order to attract sentence stress. This leads to a more careful pronunciation of the vowels and gives rise to more peripheral formant values than in spontaneous speech. Hence, the vowel reduction observed in the FAS speaker is unlikely to be contributory to the impression of a foreign accent.

The erosion of the distinctiveness of some vowels in the FAS speaker is particularly noticeable in the close front region of the vowel space: there is hardly a qualitative difference between /i/, /y/ and /e/, and between /ɪ/, /ɛ/ and /ʏ/. This smaller distinctiveness cannot be explained by the regional accent of the speaker (Verhoeven & Van Bael, 2002): therefore, it is not unreasonable to assume that this lack of distinctiveness may have contributed to the perception of a foreign accent.

At the suprasegmental level, several dimensions were studied. First, speech rate was investigated from two perspectives, that is as speech rate and articulation rate. Speech rate is expressed as the number of syllables per second, including silent and filled pauses, while articulation rate is quantified as the number of syllables per second including filled pauses, but excluding silent pauses (Verhoeven et al., 2004). In this FAS speaker, speech rate was 3.83 syllables/second and articulation rate amounted to 4.79 syllables/second. This compares well to a control group of unimpaired native speakers of Dutch who had a speaking rate and articulation rate of 3.89 syll/sec and 4.23 syll/sec respectively (Verhoeven et al., 2004). From this, it can be concluded that this speaker's speech is generally very fluent and it is precisely the dissociation in fluency between FAS and AoS that has previously been mentioned as one of the hallmark features distinguishing both speech disorders from each other (Moen, 2000; Aronson, 1990).

The next dimension that was investigated was the speaker's speech rhythm, which was quantified by means of the pairwise variability index (PVI) proposed by Low et al. (2001). This index is based on measures of vowel durations (vocalic PVI) and the duration of the intervocalic intervals (intervocalic PVI). In this speaker, the vocalic PVI amounted to 48: this is considerably lower than 65.5, which is the reference value for Dutch suggested in Grabe & Low (2002). However, it is very close to 43.5, which is the reference value for French. This suggests that the speaker's rhythm is more French-like (syllable-timed) than Dutch (stress-timed) and this may have contributed to the impression of a French accent.

Finally, the speaker's intonation was investigated along the same lines as Verhoeven & Mariën (2010a). As far as the mean pitch and the excursion sizes of the pitch movements in the contours are concerned, it was found that the speaker's mean pitch is 110.5 Hz while his pitch range amounts to 5.85 semi-tones. This agrees rather well with averages for male native speakers of Dutch suggested in 't Hart et al. (1990). The internal composition of the pitch contours was analyzed by means of the stylization method proposed by 't Hart et al. (1990). This method uses speech analysis and synthesis techniques to replace the original F0 contours by means of a minimal combination of straight lines which are perceptually equivalent. This method eliminates microprosodic variation and provides an insight in the internal structure of pitch contours. For more

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information about the application of this method to the analysis of speech pathology the interested reader is referred to Verhoeven & Mariën (2010b).

Application of the stylization method revealed 4 different pitch contours. The first one consists of a prominence-lending rising pitch movement (symbolized as 1) immediately followed by a prominence-lending fall (symbolized as A) in the same syllable. This (1-A) pattern occurred 49 times (36.6 %) in the patient's speech sample and it was always correctly associated with the most prominent syllable in the utterance. The second contour is one in which the rising and falling pitch movements 1 and A are aligned with two different prominent syllables: the two movements are connected by means of a stretch of high pitch. The occurrence of this contour is confined to the last two prominent syllables in sentences. This contour was used 13 times (9.7 %) by the speaker: all instances were well-formed and agreed with the distributional restrictions of this contour. The third contour is another variant of 1-A in which the first sentence accent is realized by means of a prominence-lending rising pitch movement (1) and the last accent is marked by means of a prominence-lending falling pitch movement (A). Any intervening accents are marked by means of a half fall (symbolized as E) and this gives rise to a typical terrace contour. The speaker used this contour 8 times (6%). The fourth contour is a continuation contour in which the accent is realized by means of a prominence-lending rising pitch movement. The pitch remains high and is then reset to a lower level in order to mark a syntactic boundary (symbolized as B). This is the standard continuation contour, which indicates that the utterance is not finished yet. This contour was used 64 times (47.8%). The 1-B contour did not always coincide with syntactic boundaries, but it was noticed that often individual words within a larger syntactic unit were realized with this contour.

The frequencies of the contours in this speech sample were compared to reference frequencies for spontaneous Dutch reported in Blaauw (1995), who carried out a perceptual analysis of instruction dialogues in 5 speakers. This comparison revealed that the frequency of occurrence of all the speaker's contours was very similar to the reference values suggested in Blaauw (1995), except for the 1B contour, which was significantly more frequent than in unimpaired speech. A similar observation was reported in Verhoeven & Mariën (2010a) and Kuschmann (2010) for neurogenic acquired FAS.

4.4. DISCUSSION

4.4.1. Semiological resemblances between FAS and DAS

This patient presented with isolated developmental motor speech problems consistent with a diagnosis of FAS and DAS. Previous research had shown that FAS can result from a compensation strategy by patients showing apraxia-like features in speech production (Whiteside & Varley, 1998). It is argued that the same can be assumed for DAS patients. Fluency has been mentioned as one of the key characteristics distinguishing AoS (Van

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der Merwe, 2009) and FAS patients, and it seems that this is semiological distinction also holds for DAS patients. Furthermore, DAS (and AoS) is often characterized by attainment of phonological sequences, whereas FAS is characterized by deviations of individual speech sounds (Moen, 2000).

This patient demonstrated many of the key features associated with DAS (Shriberg et al., 1997 (a, b); Morgan & Vogel, 2009; McCauley & Strand, 2008; Nijland et al., 2003; Terband et al., 2009; Peter & Stoel-Gammon, 2005) (see also: *neuropsychological investigations*). Some of these errors are typical segmental errors which have also been observed in other FAS cases. However, this patient did not show the typical 'trial-and-error' behavior that is regularly noted in DAS patients (Moen, 2000; 2006; Hall et al., 2007; Ozanne, 2005; Stackhouse, 1992; Terband et al., 2011). The analysis of suprasegmental features for this case provided supplementary evidence against the idea that FAS is primarily a prosodic deficit: the only remarkable feature was a syllable-timed speech rhythm and the excessive use of the 1B (continuation) contour. Speech and articulation rate, mean pitch (parameter of intonation) and the general shape of the intonation contours were normal.

4.4.2. Planning deficits: crossing speech boundaries

The hypothesis of FAS as a subtype of AoS, has previously been described from a physiological (Moen, 2000) and a cognitive perspective (Whiteside & Varley, 1998). This patient was also investigated from both perspectives. Cognitive assessment demonstrated (selective) executive disturbances (deviant scores on the Wisconsin Card Sorting Test and low results on the Stroop Task - card III) and distorted planning and organization in the visuospatial domain. However, the patient obtained average to above-average results on other executive tasks (such as the digit span and TMT-B, for instance). Comparison with the cognitive profile of the previously published cases of developmental FAS revealed a comparable discrepancy. The neuropsychological test results of the first patient published by Mariën et al. (2009) demonstrated a low average performance IQ as well as depressed scores for digit span and TMT-A and B. Scores for the WCST, Stroop task on the other hand, were well within the normal range. In their second patient, only severe syntactic deficits affecting language processing were retained. All other cognitive test results were in the average range or above. The results were consistent with a diagnosis of SLI of the phonological-syntactic type. Both the results of this patient and the first patient described by Mariën et al. (2009) go against the finding that WCST scores are a predictor for TMT-B performance, claiming that both tests give expression to attention set-shifting problems (Sánchez-Cubillo et al., 2009). Some studies have claimed that correlations between the Stroop interference and TMT-B constitute evidence of a shared expression of inhibitory control (Chaytor et al., 2006). Other studies have contradicted such a correlation. For instance, Sánchez-Cubillo et al. (2009) analyzed 41 Spanish-speaking healthy participants and found that TMT-A scores primarily tap visuo-perceptual abilities and visual search (a significant amount of the vari-

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ance in multiple regression analysis was predicted by the WAIS-III Digit Symbol score), whereas the TMT-B was primarily informed by working memory and only then by task-switching ability (their correlation with the Stroop Interference Task was nulled in the multiple regression analysis).

Functional neuroimaging with SPECT in this patient revealed a decreased perfusion in the anatomo-clinically suspected brain regions involving the bilateral prefrontal cortex, the medial frontal regions and the cerebellum. On the basis of lesion studies research has linked damage affecting the prefrontal cortex (PFC) to impaired executive functioning (Robinson et al., 1980; Yuan & Raz, 2014). Yuan & Raz (2014) carried out a literature survey about the anatomo-functional correlates of executive functions and showed that increased PFC volume in healthy subjects correlated (positively) with scores on the WCST. Buchsbaum et al. (2005) also found that perfusion in the bilateral PFC significantly increases during performance of tasks requiring executive planning and control. However, the value of the WCST as an exclusive indicator of frontal dysfunction remains a matter of debate. Chase-Carmichael et al. (1999) for instance, have contested the value of the WCST as an indicator of frontal pathology in a paediatric population (age 8–18). For their study, they classified children according to the affected brain area(s) (left hemisphere, right hemisphere, or bilateral frontal, extrafrontal, or multifocal/diffuse regions of brain dysfunction) regardless of the etiology (stroke, brain trauma, tumor, seizures, neurofibromatosis, lupus, myelomeningocele and cognitive changes of unknown origin). Results did not support the assumption that WCST performance is more impaired in frontal lesions than extrafrontal or multifocal/diffuse lesions. However, they classified all patients with frontal lobe dysfunction together and did not take into consideration differences in the affected *sub*-regions. However, they argue that dysfunction in certain sub-regions (e.g. medial frontal regions) of the frontal lobe in the left hemisphere leads to lower performance on the WCST (Grafman et al., 1986; Drewe, 1974). Still, their study confirmed that patients with left-hemisphere damage generally perform weaker than patients with right hemisphere damage. For adult stroke patients, the same conclusion holds (Jodzio & Biechowska, 2010).

This patient also obtained borderline scores on the motor integration and coordination subtests of the Beery-Buktenica Test of Visual Motor; which is a test administered to evaluate the integration of visual perception and co-ordination of fine motor skills in drawing (Beery, 1989). The patient also obtained a low score on the reproduction of the Rey Complex Figure (28/36). It was concluded from these results that the patient had spatial planning and visual structuring problems. The patient was diagnosed with a constructional dyspraxia following execution and planning problems of frontal origin.

Because visuo-constructional (Block Design, Visual Puzzles) and perceptual skills were not impaired (visual perception subtest of Beery-Buktenica), it is hypothesized that the main deficit occurs in the programming phase of the relevant motor movements prior to execution of grapho-motor tasks (Del Giudice et al., 2000). According to the model proposed by Grossi & Angelini (Grossi, 1991, see also: Grossi & Trojano,

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1998) the copying of drawings requires (1) a *visuospatial analysis* of the geometrical and spatial aspects of the figure to be copied, as well as a scan of the repertoire of internalized figures drawn in the past, (2) the *formulation of a drawing plan*, stored in the working memory (visuospatial sketchpad, Baddeley & Hitch, 1974) containing the integration of visuospatial representations into the required motor actions (programming phase) (3) the *execution of the grapho-motor movements* (4) and finally the *control of these movements* (see also: Denes & Pizzamiglio, 1999). Since this patient obtained a maximum score on the retention of visual material during neuropsychological testing, it is plausible that the impairment is situated *after* the instauration of the figure in the visuospatial sketchpad (working memory). This model is developed along the same lines as the speech sensorimotor control models (Van der Merwe, 2009). In short, the problem might be situated in the second phase of planning and programming. Furthermore, this patient did not demonstrate a hypoperfusion in the (superior) parietal region, where graphomotor plans are stored. Yet a significant hypoperfusion was found in the area circumscribing the (bilateral) prefrontal cortex, the area where graphomotor plans are programmed/integrated for execution (Mariën et al., 2013). Disorders of skilled movements, as well as underdeveloped constructional abilities have been noted in the context of DAS (Maassen 2002, Yoss & Darley, 1974; McLaughlin & Kriegsmann, 1980).

4.4.3. The hypothesis of a cortico-cerebellar network dysfunction

The frontal executive dysfunctions in conjunction with the SPECT findings lead to the hypothesis that the pattern of hypoperfusions reflect significant involvement of the cerebro-cerebellar functional connectivity network (Mariën et al., 2013; Mariën et al., 2006; Mariën et al., 2007, Meister et al., 2003, Moreno-Torres et al., 2013). Cerebellar involvement in speech disorders, including FAS and AoS, has previously been proposed from the viewpoint of the cerebellum as a coordinator of speech timing (see also: De Smet et al., 2007). Also, the phonetic analysis of our patient's speech gave evidence for semiological resemblances between DAS and FAS. However, one of the most striking differences between both conditions, namely the fluency aspect was equally confirmed for our patient. These findings provide support for the hypothesis that FAS may be a mild subtype of AoS as well the developmental cognate (Whiteside & Varley, 1998; Fridriksson et al., 2005; Mariën et al., 2006; Mariën et al., 2009; Kanjee et al., 2010; Moen, 2000; Moen, 2006).

In hindsight, diffusion tensor imaging (DTI) might be of added value to identify structural changes to the white matter tracts that make up and connect with the cortico-cerebellar tract. DTI voxel-based morphometry was unfortunately not carried out in this patient. However, it could help to further clarify the pathophysiological substrate of neurodevelopmental disorders and should be considered in future research on developmental FAS.

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4.5. CONCLUDING REMARKS

A new case of developmental FAS with DAS and a visuospatial planning disorder was presented. From a semiological as well as structural and physiological point of view, the hypothesis of a connection between FAS and DAS seems plausible in this case. Moreover, the conjunction between the speech impairment and frontal executive deficits, supported by SPECT findings provide further evidence for a potentially primary role of the cerebro-cerebellar network in both disorders. However, one of the main characteristics of DAS is trial-and-error behavior. This was not attested since the patient could adequately self-correct whenever production errors were made. Therefore, the hypothesis is put forward that FAS is a *mild* subtype of AoS, even when both are developmental in nature.

4.6. ACKNOWLEDGMENTS

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PART III: PSYCHOGENIC FAS

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CHAPTER 5

Psychogenic FAS: A Review[12]

[12] The study reported in this chapter was published in Frontiers in Human Neuroscience: Keulen, S., Verhoeven, J., De Witte, E., De Page, L., Bastiaanse, R., & Mariën, P.(2016). Foreign accent syndrome as a psychogenic disorder: A review. *Frontiers in Human Neuroscience, 10*, 1-16.

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ABSTRACT

In the majority of cases published between 1907 and 2014, FAS is due to a neurogenic etiology. Only a few reports about FAS with an assumed psychogenic origin have been published. The present article discusses the findings of a careful database search on psychogenic FAS. This review may be particularly relevant as it is the first to analyze the salient features of psychogenic FAS cases to date. This article hopes to pave the way for the view that psychogenic FAS is a cognate of neurogenic FAS. It is felt that this variant of FAS may have been underreported, as most of the psychogenic cases have been published after the turn of the century. This review may facilitate the diagnosis of the syndrome in clinical practice and it highlights the importance of recognizing psychogenic FAS as an independent taxonomic entity.

5.1. INTRODUCTION

It has now been over a century that researchers have reported on a motor speech disorder most frequently referred to as 'Foreign Accent Syndrome' (FAS). The first patient with FAS was anecdotally described by Pierre Marie in 1907. The term 'FAS' was later coined by Whitaker (1982) who also proposed a set of diagnostic criteria: (1) 'the accent is considered by the patient, by acquaintances and by the investigator, to sound foreign'; (2) 'it is unlike the patient's native dialect before cerebral insult', (3) 'it is clearly related to central nervous system damage (as opposed to an hysteric reaction, if such exist)'; (4) '(t)here is no evidence in the patient's background of being a speaker of a foreign language (i.e., this is not like cases of polyglot aphasia)' (Whitaker, 1982, pp. 196 & 198). These criteria only apply to one of the three FAS subtypes in the taxonomic classification recently developed by Verhoeven & Mariën (2010a), who distinguished between a neurogenic (including a developmental subtype), a psychogenic and a mixed variant of FAS.

Psychogenic FAS is defined by Verhoeven & Mariën (2010a) as 'the variant in which the foreign accent of the patient is grounded in underlying psychological issues' (p. 601). It is also referred to as 'non-organic', 'functional' or 'psychosomatic' FAS. Aronson & Bless (1990) have expressed a clear preference for the term 'psychogenic' because this term has 'the advantage of stating positively, based on an exploration of its causes, that the […] disorder is a manifestation of psychological disequilibrium such as anxiety, depression, personality disorder or conversion reaction […]' (p. 121). In general, this 'sub-category' contains all the cases of FAS in which an organic substrate cannot be identified after careful clinical neurological, neuroradiological and/or neurophysiological examination, and for which a clear psychological factor is identified (e.g. Verhoeven et al., 2005) as well as the cases for which it is hypothesized that a disclosed organic deficiency cannot be held responsible for the FAS (e.g. Gurd et al., 2001; Van Borsel et al., 2005). The latter is not uncommon.

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According to Baumgartner (1999) several researchers in speech and language pathology have published cases in which a clear neurological impairment was identified, but the speech or voice disorder was convincingly argued to be of psychogenic origin (Tippett & Siebens, 1991; Baumgartner & Duffy 1997). Baumgartner (1999) emphasizes the importance of carefully considering the patient's medical history, meticulously interpreting the symptoms and evaluating the coherence between different observations. If medical history, onset of symptoms, symptom characteristics and their evolution, neurological examinations, neuroimaging and cognitive work-up do not unambiguously point towards a neurological disorder, an alternative interpretation should be considered.

This article presents a detailed review of FAS cases with an assumed psychogenic etiology published between 1907 and July 2014. The focus of the investigation is on the associated psychopathologies, the onset and remission of the accent, the type of accent, the segmental and suprasegmental characteristics contributing to the perception of the patient's accent as 'foreign', as well as the comorbid speech- and/or language symptoms.

The goal of this review is to analyze the main features of psychogenic FAS in order to shed more light on this taxonomic variant and facilitate the diagnosis in clinical practice.

5.2. METHODS

The available literature on (psychogenic) FAS was identified by means of regular searches in online electronic databases (*Web of Knowledge, ScienceDirect, PubMed*, *Medline, PsycINFO*), using the following keywords in Boolean search: 'foreign accent syndrome', 'FAS', 'psychogenic AND FAS', 'psychogenic AND foreign accent syndrome'. The reference sections of all relevant articles were scanned to identify additional references. All the articles between 1907 and July 2014 were included. Only original case descriptions were retained for this review, as some of the data were re-used by the same or other authors in later publications. Inclusion criteria for psychogenic FAS were: (1) the onset of a foreign accent, (2) the presence of, or indication(s) for psychological/psychiatric symptoms, (3) the absence of neurological damage that could explain the speech and/or language symptomatology

5.3. RESULTS

5.3.1. Demographic characteristics and associated psychopathologies

The initial database search resulted in a corpus of 129 articles reporting instances of FAS (regardless of the etiology). However, at least 24 cases were published twice or more. Only original case reports were included for the counts in this section. Fifteen of the 105

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(original) FAS cases published between 1907 and July 2014 matched the inclusion criteria of psychogenic FAS (see Appendix C1). The putative psychogenic FAS cases represent 14% of all published FAS cases (n=15/105). Two case reports [case 3,8] were reported twice[13]. 67% of the included patients were women (n=10/15), and 33% were men (n=5/15). The mean age of patients with assumed psychogenic FAS was 48 years and 1 month (range: 30-74 years, SD: 12 years and 9 months). Men had a mean age of 56 years and 2 months (range 30-74 years, SD: 17 years 8 months) and women 44 years and 1 month (range 32-54 years, SD: 7 years 11 months). Patient's occupation was only mentioned in a few case reports (n=5/15) [cases 3, 5, 8, 10, 12]. Education levels were never stated. Five patients were described as right-handed [cases 2, 5, 8, 11, 12]. However, handedness was only formally assessed in one case (case 5: right-handed; Edinburgh Handedness Test; Oldfield, 1971). For the remaining cases [1, 3, 4, 7, 8, 10, 13-15], handedness was not indicated. Two patients were self-proclaimed monolinguals [cases 8, 9], whereas two were definitely polyglots [case 5: Dutch-French-English, case 10: English-Spanish]. In case 5, FAS affected both Dutch and English, but French was perfect on all linguistic levels (suprasegmental, segmental, morphology, syntax). In case 10, however, it was not mentioned to what extent the patient's proficiency of Spanish was affected. As far as the psychological disorder is concerned, 33% of the cases presented with conversion disorder (n=5/15; cases 5, 9-12), 13% with schizophrenia (n=2/15) [cases 3, 6], 13% with bipolar disorder (n=2/15) [cases 7, 8], 13% with obsessive-compulsive disorder (OCD) (n=2/15) [cases 14, 15], 7% with post-traumatic neurosis (n=1/15) [case 1], and 7% with mania (n=1/15) [case 13]. In 13% of the cases, no clear psychological disorder was associated with the FAS (n=2/15) [cases 2, 4] (see Appendix C1). However, for these cases neurological and neurophysiological examinations as well as neuroimaging were regarded incompatible with a neurogenic etiology, and it was concluded that the FAS had to be non-organic in nature.

5.3.2. PHONETIC CHARACTERISTICS

Neurogenic FAS has been associated with a very diverse set of segmental and suprasegmental pronunciation characteristics, often with great inter-patient variability. While some studies primarily investigated the phonetic and acoustic characteristics of FAS, others focused on the pathophysiological substrate of the syndrome (see also Ingram et al., 1992; Kanjee et al., 2010). This dissociation equally applies to psychogenic FAS: some researchers have focused on the identification of the associated psychopathology and the link between the psychological disorder and FAS (e.g.. Reeves & Norton, 2001; Reeves et al., 2007), whereas others described the segmental and supra-

[13] The case reported by Reeves and Norton (2001) was reported again in Reeves et al. (2007; case 3) and the case reported by Poulin et al. (2007) is identical to the case reported by Roy et al. (2012, case 1). However, all the available information was used for further analyses.

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segmental transformations in speech (Verhoeven et al., 2005; Haley et al., 2010). The speech characteristics are listed in **Table 5.1**.

Table 5.1. Overview of the segmental and suprasegmental changes in the speech of assumed psychogenic FAS. Cases marked by an asterisk are cases for which formal phonetic and acoustic analyses were carried out. For the remaining cases, the characteristics were noted based on a perceptual (impressionistic) phonetic analysis.

Segmental	Case numbers	Percentage (%) of psychogenic patients for whom speech characteristics were noted
Consonants		
Substitution (manner/place/aspect)	3,4*,5*,8*,9,10*,11,12*,13*	64
Omission	2,4*,6,7,9,10*,11,13*	57
Addition	2,4*,5*	21
Cluster reduction	4*,13	14
Increased friction	2	7
Lengthening	2	7
Vowels		
Substitution	3,4*,5*,12*,13*	36
Lengthening	2,3,8*,10*,12*	36
Addition	5*,11,12*,13*	29
Fronting	5*,8*,13*	21
Monophthongization of diphthongs	2,10*,12*	21
Reduced contrast	10*,13*	14
Lenition	9,10*	14
Backing	8*,10*	14
Omission	12*	7
Shortening	9	7
Increased tenseness	10*	7
Suprasegmental	**Case numbers**	**Percentage (%) of psychogenic patients for whom speech characteristics were noted**
Abnormal intonation	3,6,7,8*,9,10*,11,12*,13*	64
Slow speech rate	5*,8*,10*,11,12*	36
Incorrect word stress	2,4*,5*,10*,11	36
Syllable-timed speech	2,4*,8*,10*,13*	36
Variable pitch	2,10*,12*	21
Hypernasality	10*,11,12*	21
Slow articulation rate (excluding pauses)	8*,12*	14
Terminal pitch rise (errors)	7,13*	14
Larger than normal F0 excursions	8*,10*	14
Excessive pausing	5*,13*	14
Fast speech rate	13*	7
Terminal pitch fall (errors)	8*	7

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All the speech characteristics in **table 5.1** have been reported for patients with neurogenic FAS as well. It seems that in patients considered as psychogenic, vowels are more often affected than consonants and this also seems to hold for neurogenic patients (Ingram et al., 1992; Miller et al., 2006; Katz et al., 2008; van der Scheer et al., 2013). Moreover, the nature of the changes is different for vowels and consonants: consonants are mainly affected by substitutions, omissions and additions, whereas errors against vowels mostly consist of substitution errors, vowel lengthening, and additions.

5.3.3. Accents associated with psychogenic FAS

Table 5.2 shows the accents associated with psychogenic FAS.

Table 5.2. Overview of the different accents associated with FAS.

Case	Pre-FAS accent	Newly developed accent
Case 1	British English	Welsh
Case 2	British English (North Yorkshire)	French
Case 3	American English	British English
Case 4	Dutch (Belgium)	'an awkward accent'
Case 5	Dutch (The Netherlands)	French
Case 6	Southern American English	Jamaican English
Case 7	American English	'European'
Case 8	Montréal French	Acadian French, French of France, or English
Case 9	Japanese	Chinese
Case 10	American English	Eastern European
Case 11	English	French/Spanish/Jamaican/Caribbean/African
Case 12	American English	Jamaican English
Case 13	American English	Caribbean English
Case 14	Standard Dutch (The Netherlands)	Regional variant of Dutch (The Netherlands)
Case 15	Regional Dutch (The Netherlands)	Standard Dutch (The Netherlands)

In 9 out of 15 cases (60%) the accent changed between geographical variants of the same language [cases 1, 3, 6, 7, 11-15]. In 9 cases (60%) the mother tongue was a variant of English (either American or British, or a regional variant) [cases 1-3, 6, 7, 10-13]. In four cases, other variables, such as pathological language mixing [case 5] and code switching [cases 3, 14, 15], might have created the impression of FAS.

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5.3.4. Onset and remission of the accent

An acute onset of FAS occurred in 7 cases [cases 3, 6-8, 13-15]. In these cases, FAS was associated with mania [case 13], bipolar disorder [cases 7,8] and obsessive-compulsive disorder [cases 14, 15]. In the patients with schizophrenia [3, 6] the accent change co-occurred simultaneously with a psychosis. The patients who did not suffer psychiatric symptoms, related the onset of their FAS to a motor vehicle accident [cases 1, 11], a 'near-accident' [case 5], possibility of MS [case 2], a whiplash trauma 9 years prior to consultation for FAS *or* after consultation of an otolaryngologist for a change of voice quality after a minor head trauma [case 4]. For some, FAS was first noticed after admission to hospital for the sudden onset of sensory and gait symptoms [cases 9, 10, 12]. In 47% of the FAS cases that were considered psychogenic, the onset of the accent was delayed in comparison to the occurrence of the adverse life event that was held responsible for the FAS by the patients themselves [cases 2, 4, 5, 9-12]. In 5 of these cases, the patients were diagnosed with a conversion disorder [cases 5, 9-12].

In 27% of the cases[14] (*n*=4/15) [cases 3, 6, 7, 13], the accent receded simultaneously with the associated psychiatric disorder. In two cases (13%) [cases 4, 10] FAS receded spontaneously. In all other patients [1, 2, 5, 8, 9, 11, 12, 14, 15], FAS remained present throughout follow-up. In case 5, scores on the Minnesota Multiphasic Personality Inventory (MMPI; Butcher et al., 1989) and Dissociation Questionnaire - Revised (DISQ-R; Vanderlinden et al., 2009) were *near* the accepted mean, but the accent persisted.

Only three patients received speech-language therapy to reduce FAS [cases 4, 10, 12]. Van Borsel et al. (2005) applied auditory masking and delayed auditory feedback (see also comments of Moreno-Torres et al., 2013). However, these interventions did not resolve FAS. Case 10 received a symptomatic intervention for psychogenic voice and speech disorders (Duffy, 2005). However, progress did not transfer to conversational speech and the accent suddenly receded after having quit outpatient therapy for several weeks. Case 12 agreed to behavioral speech therapy as well (targeting the production of individual speech segments), but she quit after one session for reasons that were not disclosed.

For patients whose accent change *receded* during follow-up [cases 3, 4, 6, 7, 10, 13], the period between accent onset and remission was about 63 days on average, i.e. 9 weeks (range: 6 days - 6 months, SD: 71 days). The patient described by Reeves & Norton (2001) [case 3], was re-admitted to hospital three times and this was taken into account for the calculation of the duration. In 60% of the cases [cases 1, 2, 5, 8, 9, 11, 12, 14, 15] the accent did *not* resolve. In these patients, investigation of the period between accent onset and last follow-up revealed that the accent persisted for 45 months on average[15] (range: 15 months - 8 years; SD: 28 months and 2 days).

[14] Case 9 and case 11 could not be included in this count due to a lack of information

[15] The exact duration is unknown. The calculated figure is entirely dependent upon the duration of the follow-up for reported case studies.

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5.3.5. Psychodiagnostic and neuropsychological testing

Table 5.3. Overview of the patients subjected to psychodiagnostic tests.

Psychodiagnostics	
Test	**Case number(s)**
MMPI-2 (Butcher et al., 1989)	5,11,12
DISQ-R (Vanderlinden et al., 2009)	5
BDI-2 (Beck et al., 1996)	12
NEO-PI-R (Costa & McCrae, 1985)	12
SCL 90-R (Derogatis, 1983)	12
STAI (Spielberger et al., 1970)	12

Abbreviations: MMPI-2= Minnesota Multiphasic Personality Inventory-II; DISQ-R= Dissociation Questionnaire Rev; BDI-2= Beck Depression Inventory-2; NEO-PI-R= Neuroticism Extroversion Openness Personality Inventory, Rev; SCL-R= Symptoms Checklist-90-items, Rev; STAI= State Trait Anxiety Inventory.

Formal psychodiagnostic testing was carried out in three patients (see **Table 5.3**). In case 5, the results obtained on the MMPI-2 in 1995 showed a conversion V-pattern. The conversion V-form designates a markedly low score on the depression scale (scale D): the conversion suppresses depression, which explains lower scores on scale D. On the other hand, it is associated with increased physical sensations, thereby increasing scores on the hypochondriasis scale and hysteria scale (Leavitt, 1985). The second patient's profile elicited an elevated degree of defensiveness (K: 70) and hysteria (Hys: 61). The restructured clinical scales revealed marginally elevated scores for depression (RC2: 66) and somatic complaints (RC1: 57). The elevated scores on the hysteria scale in conjunction with the somatic complaints (although only marginally elevated) are additional arguments to suspect conversion disorder. However, the typical V-pattern was not found. Although exact scores were not provided, a conversion-V profile was also found on the MMPI-2 for case 12 (code type 1-3/3-1 is generally associated with conversion disorder). Scores on the neuroticism scale of the NEO-PI-R were low, which indicates stable personality and emotions, calmness, but also a decreased reactiveness to everyday situations (Nelson, 2014). The patient scored in the average range for the extraversion, agreeableness and conscientiousness scales. No mention was made of scores for openness to experience. The SCL-90-R is a "90-item self-report symptom inventory" (Derogatis & Savitz, 1999) in which the patient rates the severity of a series of psychiatric symptoms. These are grouped around nine dimensions: somatization, obsessive-compulsiveness, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation and psychoticism (Domino & Domino, 2006). Only one clinical score was mentioned, i.e. for the somatization scale (T= 65). This agrees well with the

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profile elicited on the MMPI-2. The STAI is a self-report scale for anxiety consisting of two 20-item scales. The patient indicates (1) how he/she feels now (*state*) and (2) how he/she feels generally (*trait*) (Lam et al., 2005). Scores on the STAI were subclinical. Finally, the BDI-2 is a self-report inventory, which consists of a series of statements concerning complaints. The patient notes how he/she feels about the statements taking into account his/her psychological status over the last week. Scores on the BDI-2 were equally sub-clinical.

Only in a small number of case studies formal neuropsychological investigations were carried out. General cognition, memory, attention, executive functioning, *and* language were assessed in 5 cases [cases 2, 5, 8, 11, 12] (see **Table 5.4**).

Table 5.4. Overview of the patients subjected to neuropsychological tests.

Neuropsychology	
Test	**Case number(s)**
General cognitive screening tests	
MMSE (Folstein et al., 1975),	5
CLQT (Helm-Estabrooks, 2001)	10
WRAT (Wilkerson, 1993)	11*,12
Intelligence	
WAIS (Wechsler, 1981, 1997)	5,9,11,12
Memory	
WMS (Wechsler, 1991, 1997),	5,12
Brown Peterson Task	8
CVLT (+learning) (Delis et al., 2000)	12
RAVLT (+learning) (Rey, 1941)	11
BVMT-R (Benedict, 1997)	12
Attention, set-shifting	
Stroop task (Stroop, 1935)	5,8,11,12
Ruff figural fluency (Ruff, 1988)	12
TMT (Reitan, 1958; 1992)	5,8,11,12
Visuospatial abilities	
Rey complex figure (Rey, 1941)	5,11
Judgment of line orientation (Benton et al., 1983)	5
Motor functioning	
Finger tapping test (Arnold et al., 2005)	11,12
Grooved pegboard (Klove, 1963; Lafayette Instrument, 2002)	12

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Symptom validity tests	
Green word memory test (Green, 2005)	12
Dementia scales	
HDS (Cole et al., 1983)	5
ADAS (Rosen et al., 1984)	5
Language	
BNT (Kaplan et al., 2001)	3,6,7,10-12
PPTT (Howard & Patterson, 1992)	8
Token Test (De Renzi & Vignolo, 1962)	4,8,12
BDAE (Goodglass et al., 2000).	2*,3,5,10
AAT (Graetz et al., 1992: Dutch version)	4*,5
MAE (Benton et al., 2001)	10*,11*,12*
SAN-TEST (Deelman et al., 1981)	4*
DO-80 (Deloche & Hannequin, 1997)	8
Picture naming via an experimental test	2
PENO (Joanette et al., 1990).	8*
Phonemic fluency (FAS) (Norms: Tombaugh et al., 1999, case 11; Benton et al., 2001: case 12; case 5: unpublished norms)	5,11
Semantic fluency (animals, transport, vegetables, clothes: unpublished norms)	5
Word/sentence reading via an experimental test	2
Word sentence spelling via an experimental test	2

2*: possibly only two subtasks of the BDAE were administered: the non-verbal and the verbal agility test.
4*: only written language via AAT; sentence comprehension and word retrieval (animals) SAN-Test
10*: auditory word and sentence comprehension, sentence repetition, and oral and written spelling MAE.
11*: word reading and spelling tests of the WRAT; sentence repetition task, as well as the aural and reading comprehension task MAE
12*: repetition skills, auditory comprehension, token task, and reading comprehension MAE.

Abbreviations: MMSE= Mini Mental State Examination, WAIS= Wechsler Adult Intelligence Scale, WMS= Wechsler Memory Scale, TMT= Trail Making Test, WRAT= Wide Range Achievement Test, CVLT= California Verbal Learning Test, RAVLT= Rey Auditory Verbal Learning Test, CLQT= Cognitive Linguistic Quick Test, BVMT-R= Brief Visuospatial Memory Test-Rev, HDS= Hierarchic Dementia Scale, ADAS= Alzheimer's Disease Assessment Scale, BNT= Boston Naming Test, PPTT= Pyramid and Palm Tree

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Test, MAE= Multilingual Aphasia Examination, BDAE= Boston Diagnostic Aphasia Examination, AAT=Akense Afasie Test (Dutch version), SAN-test= Stichting Afasie Nederland-test, DO-80= Test de Dénomination Orale d'Images, PENO= Protocole d'Evaluation Neuropsychologique Optimal

In case 9, only intelligence was investigated. In cases 3, 4, 6, 7, and 10 only language testing was performed. Neuropsychological examination consisted of a variety of tests (**Table 5.4)**.

Cognitive performance was 'within normal limits' (p. 715, Gurd et al., 2001) for case 2 and average to above average on all tasks in case 5. In case 8, memory and attention were normal, but the patient gave evidence of difficulties with short-term memory (Brown Peterson Task: mean of interference scores: 42%; norm: 97.22%, SD: 4.46), as well as with attention control and executive functions (Stroop test: Stroop effect: 249", norm: 142.4", range: 88–204"; TMT-A: 61", norm: 41.3", SD: 15" and TMT-B: 253", norm: 111.4", SD: 72.2"). In case 9, results on the WAIS-R were within the normal range (VIQ=96, PIQ=107 and FSIQ=101). Case 11 had poor executive functions (Stroop test, Interference <1 pc., and TMT-B: 83", mean: 56.0, SD: 21.2), problems with attention and poor processing speed (TMT-A: 43", mean: 23.8, SD: 6.9, Stroop test A: 101", <1 pc.). Case 12 demonstrated impaired intelligence, memory, attention, executive functions and fine-motor skills: WAIS-III (FSIQ= 65, VIQ= 76, PIQ= 60); Trail Making Test (146"), Grooved Pegboard (dominant hand: 149", mean=85", range: 48"-121", non-dominant hand: 130", mean=101"; range: 47-152"), and Green Word Memory Test (Green Word Memory Test: immediate= 87.5, delayed= 77.5, consistency= 70.0).

Most patients in whom language was assessed, obtained average to above average results [cases 3-7, 10]. Case 2, however, had impaired oral agility as demonstrated by the BDAE (non-verbal agility: 4/12 and verbal agility: 7/12). Case 8 presented with (severely) depressed scores on phonemic and semantic category fluency (letter fluency: 5, mean: 45.46, SD: 16.4; category fluency: 14, mean: 47.85, SD: 9.8). Case 11 obtained depressed scores on most tasks evaluating speech and language (WRAT; reading: 43, pc. 6; spelling: 43, pc. 37); MAE sentence repetition (A: 2, < pc. 1 and B: 3, < pc. 1), verbal fluency (FAS): 19, pc. 2. Case 12, also demonstrated low average to impaired scores on most of the administered tasks: the BNT score was considered low average (41/60). On the MAE the following scores were obtained: repetition: 5 (impaired); auditory comprehension: 15 (borderline impaired), token test (as part of MAE): 40 (low average), and reading comprehension: 16 (borderline).

5.3.6. Comorbid speech and language disorders

Five cases suffered from additional speech and/or language deficits [cases 4, 5, 8, 11, 12], apart from FAS. Case 4 (Van Borsel et al., 2005) and case 12 (Jones et al., 2011) went through a period of pre-FAS mutism. In case 4 mutism was only documented by self-report. Van Borsel et al. (2005) noted that the patient's language was characterized by grammati-

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cal anomalies. This was also the case for the patient of Poulin et al. (2007) [case 8].

Case 5 implemented French syntax in native Dutch speech. Non-fluent expressive output was characterized by mistakes typically made by French learners of Dutch. Oral output of case 11 was initially considered as dysarthria, later as 'apraxia of speech' (p. 1010). As mentioned, the patient obtained lower scores for verbal fluency (F,A,S), but also for sentence repetition (MAE A&B: pc. <1) and the reading and spelling tasks of the WRAT (reading: 43, pc. 6; spelling: 43, pc. 37). It is expected that these symptoms are related to neurological damage. Indeed, apraxia of speech is caused by structural damage to the anterior insula of the language dominant hemisphere (Dronkers, 1996). Nevertheless, contrary to expectations, repeat structural imaging of the brain (CT and MRI) did not disclose any damage. In addition, FAS was accompanied by 'telegraphic speech' (irregularly deleting prepositions, for instance). In this particular case, the comorbid symptoms and the language deficits were regarded as 'not credible' because the extent of the deficit did not correspond to neuroimaging findings. The patient was diagnosed with FAS of a non-organic nature because of inconsistencies in the language symptoms.

5.4. DISCUSSION

5.4.1. Demographic data

Analysis of the available literature suggests that psychogenic FAS is quite rare (*n*=15/105) (14%). During the past decade FAS has increasingly attracted the attention of the scientific community as 93% of the psychogenic FAS cases (*n*=14/15) were published in a time span of only twelve years (2001-2013). The finding that there are more women with psychogenic FAS than men (67% are women, 33% are men), might be partly explained by the increased predisposition of women to several of the associated psychopathologies. Most mental disorders are also more prevalent among women than men (see also: World Health Organization, 2014). For schizophrenia, prevalence figures are esteemed to be equal, irrespective of gender, although symptoms occur earlier in men (Angermeyer & Kühnz, 1988; Saha et al., 2005; National Institute of Mental Health, 2015). On the other hand, the analysis of the neurogenic population revealed a similar demographic distribution: 68.6% of the authentic (neurogenic) FAS cases were women (*n*=59/86). Interestingly, Baker (2003) points out that it should also be taken into account that women are twice as likely to seek medical attention than men. It thus seems that the explanation for this demographic distribution remains speculative.

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5.4.2. Associated psychopathologies

Several different psychopathologies have been associated with FAS. In patients with schizophrenia, all FAS episodes *co-occurred* with a discontinuation of anti-psychotic drugs, which caused exacerbations [cases 3, 6]. In the bipolar patients FAS also co-occurred with positive symptoms [cases 7, 8]. Reeves et al. (2007) put forward the hypothesis of a direct link between the manic/psychotic exacerbations and FAS in their patients via a Positive And Negative Syndrome Scale (PANSS; Kay et al., 1987). They also suggested that FAS could have been related to a temporary disruption of the inhibition of the bilateral superior temporal gyri (STG) during exacerbations. The STG is inhibited in healthy controls when the left dorsolateral PFC is activated for word generation. It is hypothesized that FAS may have been caused by the intermittent suppressed neural circuitry.

Moreno-Torres et al. (2013) observed that the dopaminergic system may be disrupted in FAS patients. The intake of dopamine antagonists (olanzapine, risperidone) in case 3 and 6 could have restored the neurotransmitter balance and diminished the FAS. Particularly in schizophrenic patients, the so-called 'dopaminergic hypothesis' (Meltzer & Stahl, 1976; McCutcheon & Stone, 2015) agrees well with this theory. This hypothesis claims that positive symptoms in schizophrenia can be reduced by the intake of dopamine antagonists or dopamine D2-receptor blockers. It has also been shown that modulation of the dopaminergic system influences the functioning of the (pre) fronto-striato-pallidal-thalamic network, which is hypothesized by Reeves & Norton (2001) to be implicated in the accent change, and has been related to the occurrence of psychosis (Honey et al., 2003).

The symptoms of case 13 might be explained along the same lines, as excess dopamine transmission has been suspected to incite manic symptoms (Swerdlow & Koob, 1987; Cookson, 2013). Nevertheless, the pathophysiology of both psychiatric disorders is characterized by subtle differences. In schizophrenia, abnormal activity occurs in the striatum and the prefrontal cortex, whereas in mania the activity may be located more towards the dorsal nigrostriatal pathways (Cookson, 2013). Nevertheless, Cookson (2013) reported that antipsychotic drugs such as risperidone, and olanzapine (dopamine antagonists, and more specifically the ones administered to the schizophrenic FAS cases: case 3 and 6) work well on manic symptoms, such as pressured speech. The speech of case 13 was marked by excessive pressure, increased speed, loudness and forcefulness. The patient's FAS receded simultaneously with resolution of mania after pharmacological treatment.

In case 8, a psychiatrist related the accent change and sudden Spanish and German sounding words to a psychological problem at a subconscious level. Poulin et al. (2007) performed a ^{18}F-FDG-PET scan which demonstrated metabolic changes in the area of the left insular and anterior temporal cortex and a diffuse hypoperfusion affecting the frontal, parietal and temporal lobes bilaterally. MRI of the brain showed a slight asymmetrical atrophy. All imaging was performed in euthymic state. The possibility that both the language and psychological disorder were consistent with the neuroradiologi-

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cal findings was considered. However, the alterations at a linguistic level remain odd, even in the light of the attested neuroradiological findings. For instance, the output of the patient – contrary to what is expected in cases of agrammatism – was fluent, and despite a hypoperfusion affecting the insula, articulation was perceived as normal in every respect. There was no sign of apraxia of speech-, dysarthria- or aphasic-like symptoms. All of the investigated linguistic functions were normal, except for a deficit in letter and category fluency.

Case 14 and 15 suffered from refractory OCD and were treated by means of deep brain stimulation (DBS). They both developed hypomanic behavior and started experiencing accent changes afterwards. The hypothesis of FAS due to an undetected lesion induced by the electrode implantation was excluded, as the accent only developed *after* the actual stimulation by the electrode and post-operative CT confirmed the absence of any additional structural brain damage. Furthermore, Polak et al. (2013) argue that lesions caused by DBS are smaller than those generally associated with FAS, including the peri-sylvian area, (pre-)motor area, and insula of the language dominant hemisphere. However, dysfunction of the previously mentioned cortico-striato-pallidal-thalamic loop has frequently been suspected to be the pathogenic mechanism behind OCD, and the function of this circuit is altered when the nucleus accumbens is targeted for DBS.

'Hysteria,' or 'hysteric reaction', the term Whitaker (1982) used as an exclusion criterion for FAS, is an outdated term for 'conversion disorder' [cases 5, 9-12]. Conversion disorder has been subsumed under the concept of '*hysterical* neuroses' in the DSM-II (American Psychiatric Association [APA], 1968). According to Aronson & Bless (2009) a conversion reaction can affect any system requiring sensory or voluntary motor control and hence, also voice and speech. DSM-IV-TR (APA, 2000) criteria allow for such an interpretation as well, although the concept has frequently been the object of debate and is regarded insufficiently clearly defined to allow for a conclusive diagnosis (e.g. Delis & Wetter, 2007; Stone et al., 2011). In all psychogenic FAS patients with conversion disorder or those patients for whom the hypothesis of a conversion disorder was raised, the shift in accent was never the 'first' conversion symptom to occur: all case studies report more general physical discomforts that *preceded* the FAS. Especially gait and balance disturbance [cases 5, 9, 10-12] occurred but also a range of sensory problems including tinnitus [case 9], left-sided weakness affecting face and arm [case 10], blurred vision [case 10], altered hearing [case 10], abnormal sensations in arms and legs [case 10], facial numbness [case 11], weakness in the right arm [case 11], deafness to the left ear [case 11], give-way weakness [case 12], and a right-side sensory loss [case 12].

In cases 2 and 4 an associated psychological disorder was not obvious, rather there was a range of clinical observations and findings from radiological and neurophysiological investigations, which suggested a potential psychogenic origin of FAS. Gurd et al.'s patient (2001) [2] was qualified as 'psychogenic', even though CSF analyses revealed oligoclonal bands, a bio-marker of Multiple Sclerosis (MS) and EEG revealed transient spikes over the left temporal lobe. T2 hyper-intensities were found on MRI (judged clinically insignificant). It is therefore questionable whether patients suffering from MS

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(Gurd et al., 2001, Villaverde- González et al., 2003, Bakker et al., 2004, Chanson et al., 2009) really develop FAS as a consequence of their neurological disorder or due to accompanying psychological distress. Grazioli et al. (2008) note that over 50% of the MS patients suffer from depression. Case 2 obtained borderline results on the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983). The case of Bakker et al. (2004) was noted to have very 'labile emotions' (p. 271). The case of Villaverde-González et al. (2003) had a history of depression as well as an elevated irritability (p.1035). For the other patients, psychological well-being was not indicated.

Van Borsel et al.'s (2005) patient [case 4] had no demonstrable lesions on CT, and displayed no symptoms apart from a change of accent and some articulatory and grammatical difficulties. She had sustained a head trauma and whiplash nine years earlier and had suffered from chronic headaches ever since. Her accent change had occurred after a visit to the otolaryngologist, approximately one month after she had suffered another minor head trauma. Van Borsel et al. (2005) diagnosed the speech disorder as non-organic FAS because of a psychiatric history (depression and suicidal ideation) which was related to marital problems, a completely normal neurolinguistic assessment apart from mild grammatical anomalies, articulatory difficulties and an accent change, the absence of a organic deficit, and a spontaneous resolution of the accent five months after the initial visit.

Case 11 suffered a minor head trauma as well but developed FAS only three years later, associated with intermittent, atypical expressive language deficits, and apraxic as well as dysarthric symptoms. Initially, she also claimed that she was deaf to her left ear, but a hearing loss was formally ruled out. The patient displayed an 'inconsistent' agrammatism, characterized by deletions of function words. She would use and subsequently erase the same words in a series of successive utterances. She also made other inconceivable mistakes, such as splitting numbers into digits. Given the high degree of automaticity of such numerical output, these errors are highly unlikely to occur in the absence of other language deficits. Since she passed most of the symptom validity tests, she was considered not to be feigning or malingering and was ultimately diagnosed with conversion disorder.

5.4.3. Segmental and suprasegmental characteristics

Patients with FAS of an assumed psychogenic etiology present with a variety of segmental and suprasegmental errors. At the segmental level, the image more or less corresponds to what is generally found in neurogenic patients, including a dissociation between vowels and consonants (e.g. Katz et al., 2008). At the suprasegmental level, slow speech rate is often seen [cases 5, 8, 10-12]. Slow speech rate can be linked to slow processing speed, which may occur as a consequence of psychological and psychiatric impairment (e.g. depression, post-traumatic stress disorder, bipolar disorder, and schizophrenia). Analysis of (psychogenic) FAS-related segmental and suprasegmental errors has been predominantly impressionistic, except for a few cases in which (acoustic) measure-

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ments (e.g. fundamental frequency, speech intensity, speech and articulation rate) were also included [cases 5, 8, 10, 12, 13]. Deviant intonation [cases 3, 6-13] is a function of pitch variation. Intonation was aberrant in most patients with a reduced speech rate [cases 8, 10-12], but also in patients who spoke at a normal or even fast pace [case 13]. In four cases [cases 3, 6, 7, 13], deviant intonation may be associated with a psychopathology. In schizophrenia [cases 3,6], difficulties with receptive-affective prosody have been described (Rossell et al., 2013). However, Hoekert et al. (2007) state that dysfunctional *expressive* affective prosody also qualifies the speech profile. The manic patient of Lewis et al. (2013) demonstrated fast speech (FAS: 229 wpm; base line speech [BL]: 173.9 wpm; average speech rate: 190 wpm based on Yorkston et al., 1996) and a pitch level that was considerably higher during FAS than during the baseline condition (conversational speech; FAS: 265.63 Hz, BL: 160.56 Hz; average F0 for a woman: 160-225 Hz based on Baken, 1987; Titze, 1994) (see also: Hanwella & de Silva, 2011). A higher speech rate was negatively correlated with the size of the vowel space, i.e. a higher speech rate leads to a more compressed vowel space in non-brain damaged subjects, which was exactly what Lewis et al. (2013) found in their patient. This compression could explain the reduced intelligibility of speech in comparison to the BL conversation sample (FAS: 73% vs. BL: 100% intelligible): contrasts between vowels diminish and vowel duration is shortened (Chen et al., 1983; Turner et al., 1995; Weinrich & Simpson, 2014).

5.4.4. Accent change

The overview of the different accents of the analyzed cases shows that there does not seem to be any consistency. However, some interesting observations can be made. Firstly, it is striking that in 7 out of 15 cases (47%) the accent changed from the standard language variant to a regional one, or the other way round. In 9 cases (60%) the mother tongue was some variant of English: either British English [cases 1, 2] or American English [cases 3, 6, 7, 10-13]. FAS is frequently documented in Anglo-saxon media[16], as such the syndrome is more commonly known among layman. For some cases more than *just* the accent gave the listeners the impression of a very specific foreign accent: language mixing (e.g. case 6) and code switching [case 3, 14, 15] were also observed. Code switching can be defined as switching between language varieties or registers within a single conversation. For case 3, this involved the use of words such as '*blokes'* instead of the usual American variant '*friend*'. Case 14 occasionally[17] used a dialectal

[16]- Madlen, Davies, "The woman with Foreign Accent Syndrome: Mother goes to bed with broad Staffordshire accent and wakes up sounding POLISH", *MailOnline*, October 2nd 2014, accessed on March 23rd, 2015, http://www.dailymail.co.uk/health/article-2778297/The-woman-Foreign-Accent-Syndrome-Mother-goes-bed-broad-Staffordshire-accent-wakes-sounding-POLISH.html;

- "Embarrasing bodies, Conditions: Foreign Accent Syndrome," *channel4embarrassingillnesses.com*, accessed on February 2nd, 2015; http://www.channel4embarrassingillnesses.com/conditions/foreign-accent-syndrome/

- Thomas, Emily, 'Sarah Colwill Speaks Out About Foreign Accent Syndrome In BBC Documentary "The Woman Who Woke Up Chinese"', *Huffingtonpost.com*, April 4th, 2013; accessed on 23rd March, 2015; http://www.huffingtonpost.com/2013/09/04/sarah-colwill-_n_3869077.html

[17] no examples were provided

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variant of Dutch while case 15 employed vocabulary typical of a more formal register. The patient used words such as '*public toilet*' instead of the more informal: '*loo*'. Polak et al.'s (2013) patients' alterations could be related to DBS, as such linguistic modifications can occur after stimulation. Verhoeven et al.'s (2005) 51-year-old female patient (case 5) occasionally used French words, made literal translations from French to Dutch, and adapted syntactic structures resembling the Dutch of second language learners. It has to be mentioned that this patient had been a teacher of Dutch in a French company based in Holland and this may have rendered her very conscious of mistakes generally made by French learners of Dutch. These symptoms constitute another point of difference between the neurogenic and psychogenic patient population, as the insertion of foreign words or regional expressions was previously only noted in a case of Ryalls & Whiteside (2006: insertion of British equivalents of American expressions) and a case of Laures-Gore et al. (2006, case 2: insertion of Spanish words in English speech). Both case reports, however, represent instances of mixed FAS (see also Verhoeven & Mariën, 2010a). 'Pure' neurogenic FAS patients who demonstrated such lexical excursions have not been identified.

5.4.5. Psychodiagnostic and neuropsychological testing

Only three patients were tested with formal psychodiagnostic test batteries. Only in two patients [case 5, 12] the pattern was significant for a conversion disorder. In case 11, somatization and hysteria were (slightly) elevated and a diagnosis of conversion disorder was agreed upon based on the inexplicable symptom course and the presence of symptoms that could not be explained on the basis of neurological impairment (apart from the FAS, sensory and motor problems equally occurred: see also 5.4.2). For case 9, who underwent a psychodiagnostic interview, family conflict was regarded to have had such a profound effect on the patient's mental state, that the symptoms could be related to psychological problems and a childhood trauma.

Only for case 11, additional symptom validity tests were administered. Incorporation of these tests in psychodiagnostic testing is always recommended, not only when secondary gains are at stake [case 11], but also when the impact of traumatic experiences or psychological discomforts are (possibly) downplayed (Cima et al., 2003, Bush et al., 2005). In these cases, it is important to interpret neurocognitive test results with caution, as these too can be consciously manipulated (see also: 'cogniform condition/disorder': a recently developed concept within the somatoform disorders; described by Delis & Wetter, 2007).

With respect to neuropsychological testing, results were diverse for scores on tasks evaluating memory, intelligence, executive functions and attention. Three out of the five patients diagnosed with conversion disorder had poor memory and/or attention and executive functions [cases 8, 11, 12] and in one instance, deficits in fine motor skills were also observed [case 12]. Deficits in learning and memory, but also in executive functions, attention, processing skills and word finding have been associated with so-

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matoform disorders (Niemi et al., 2002; Trivedi, 2006; Demir et al., 2013). Especially, attention and executive functions are often impaired in this patient group. One of the hypotheses that has been raised to explain cognitive impairment in this group is that these deficits relate to frontal brain dysfunction. However, Wall et al. (2013) point out that the studies that claimed an association between cognitive deficits and conversion disorder did not include symptom validity tests in their test protocol for patient selection and therefore no generalizations can be made. Still, the authors argue that the incidence of neurologically inexplicable cognitive deficits in patients with conversion disorder is quite high. It remains unclear whether there is a fixed set of neurocognitive deficits specific to this population, or, as others argue, whether the deficits are related to the associated psychiatric distress (Lamberty, 2008).

5.4.6. Remission of the FAS

In the neurogenic population a late onset of FAS has only been noted when the FAS was 'masked' by other speech or language disorders (mutism, Broca's aphasia, apraxia of speech or dysarthria). Apart from a pre-FAS muteness [cases 4, 12] and apraxic/dysarthric-like symptoms in one case [case 11], FAS was never 'masked' by preceding speech/language deficits in current group. Hence, a delayed onset might be indicative of a psychogenic origin. For 27% of the investigated patients (n=4/15), FAS receded simultaneously with the remission of the related psychopathology [cases 3, 6, 7, 13]. In those cases, FAS developed after psychosis or after a (hypo)manic attack and was associated with a sudden withdrawal of neuroleptic drugs, or an unbalanced drug intake. In two cases (13%), FAS receded spontaneously [cases 4, 10]. Only three patients received speech-language therapy in order to reduce the FAS [cases 4, 10, 12], and case 11 received speech-language therapy before the accent appeared. Case 10 received the symptomatic speech therapy as proposed by Duffy (2005). According to the authors, the patient occasionally managed to accurately realize the target items, although she herself did not embrace her progress. Delayed auditory feedback and auditory masking did not improve the speech deficits in the patient reported by Van Borsel et al. (2005), although this approach has been advocated by other researchers as well (González-Álvarez et al., 2003; Moreno-Torres et al., 2013). Butcher et al. (2007) point out that there is a lack of evidence-based treatment strategies for psychogenic speech and language disorders, and that this is directly related to the uncertainty and lack of confidence on the part of the speech therapist to diagnose a disorder of psychogenic origin. To the best of our knowledge, no large-scale study has ever been carried out to evaluate the effectiveness of a treatment for psychogenic *speech* disorders.

5.4.7. Comorbid speech and language deficits

Appendix C1 shows that two patients [cases 4, 12] were mute before the onset of FAS. Psychogenic mutism is well-recognized (Salfield, 1950; DSM-V: APA, 2013). For case 4,

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the mutism can be related to the impact of psychological issues (depression, suicidal ideation) as well as to severe anxiety problems (permanent fear that the patient's son might develop Huntington disease). Case 12 was diagnosed with a conversion disorder. Mutism has previously been diagnosed in patients with conversion disorder and, in those specific cases, it is also referred to as 'conversion mutism' (Rothbaum & Foa, 1991; Aggarwal et al., 2010).

In three cases, language was also characterized by agrammatic output [4, 8, 11]. McKenna & Oh (2005) note that Karl Kleist as early as 1914, used both the terms agrammatism (non-fluent, as in Broca-like speech; mostly seen in catatonic patients) and paragrammatism (fluent, more as in Wernicke-like speech; mostly seen in paranoid patients) in a psychiatric context. In 1976, Norman Geschwind described the case of a patient with a 'hysterical pseudo-agrammatism'. The patient had been locked up in prison for passing bad checks, after which he suddenly developed a strange speech disorder and was admitted to a mental institution. What struck Geschwind was that the patient produced agrammatic speech at a normal rate in combination with stuttering behavior, a combination of symptoms, which according to Geschwind was 'unique' (p. 81) and very unlike what is seen in agrammatic aphasic patients. In 1983, Levy & Jankovic published an experiment, in which they induced a (placebo) conversion reaction in a female patient in her mid-twenties. The researchers set up a double-dissociation experiment: first, the patient received a saline injection, but she was told it contained phenytoin. Later, she received the phenytoin injection, but this time she was told it contained 'a neutral substance'. The patient's neurological symptoms worsened after each explicitly mentioned 'raise' in phenytoin, as did her scores on the various neurolinguistic exams (among others: the BDAE; Goodglass & Kaplan, 1972). Her speech became slower, (moderately) slurred and hypophonic. She made several literal paraphasias, used a telegrammatic style in repetitions and spontaneous speech, and employed overgeneralizations in picture naming. After the medicine was told to 'have worn off' completely, neurolinguistic testing demonstrated only one (!) naming error. De Letter et al. (2012) reported three cases with (non-fluent) agrammatism, overgeneralizations and paraphasias that could not be attributed to an underlying organic cerebral pathology. All three patients presented with psychiatric conditions: case 1 suffered from bipolar disorder, case 2 had a 'manipulative personality' (p. 877) and case 3 had quite an extensive psychiatric history marked by mood swings, depression and aggressiveness. All patients produced non-fluent speech, characterized by excessively long pauses. Furthermore, the patients demonstrated hypophonia, persevered in their errors, and spoke with a reduced speech rate. As was the case for the patient of Levy & Jankovic (1983) the patients never produced frustrated reactions and never attempted self-correction. For De Letter et al. (2012) the fluctuating language problems and neurological symptoms were the primary reasons for considering the speech/language problems of their patients as psychogenic, although they demonstrated organic anomalies. They argue that "the presence of a language disorder in patients with organic cerebral disease cannot demonstrate causation (e.g., Whitlock, 1967)" (p. 876).

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Van Borsel et al. (2005) explicitly argue that "grammatical anomalies […] did not conform to the pattern of agrammatism typical of Broca's aphasia or paragrammatism as seen in Wernicke's aphasia" (p. 424). In case 8, the agrammatism was equally noted in a context of otherwise well-articulated, fluent speech. However, apart from verbal fluency deficits (category and letter fluency) in case 8, there were no other notable deficits that characterized the neurolinguistic profile of most of these agrammatic patients. For case 11, it was mentioned that the patient had an agrammatism that was typologically different from Broca-aphasia (Kean, 1977, 1985): e.g. the patient was fluent and speech was not consistently agrammatic as she was able to rephrase sentences, and use initially omitted prepositions or verbs.

The case described by Cottingham & Boone (2010) [case 11] also presented with dysarthria-like symptoms and a suspected apraxia of speech, although no structural lesions were seen on CT or MRI. Hence, the speech and language symptoms of their patient were considered as 'non-credible'. There are other reports of patients demonstrating similar incredible language symptoms. Recently, a report of De Witte & Mariën (2015) observed inexplicable post-operative language symptoms and considered them as psychogenic in a 28-year-old male patient, who had undergone awake surgery for the removal of a tumor in the left anterior inferior temporal gyrus. Post-operatively, the patient was able to repeat, read, write, name high and middle frequency words but auditory comprehension and naming of low frequency words were severely impaired and he displayed inconsistent comprehension deficits. It was noted that results on the CES-D (Center for Epidemiological Studies Depression; Eaton et al., 2004) and STAI (Spielberger et al., 1983) were higher than the cut-off, indicating a higher risk for depression or anxiety disorder. De Witte & Mariën (2015) hypothesize that the symptoms of their patient were non-organic because of the patient's sensitivity to stress and depression, the atypical (course of the) symptoms, and the fact that, despite the comprehension deficits, the patient had very good insight in the disorder as his aunt suffered from vascular aphasia. If the symptoms themselves, or the course of the symptoms, cannot be explained by attested neurological deficits, the possibility of a psychogenic etiology should at least be considered (see also: Baumgartner, 1999)

The case reported by Verhoeven et al. (2005) [case 5], presented with a form of 'pseudo-paragrammatism'. This patient's speech was characterized by mistakes typically made by French learners of Dutch. The patient did not speak in a telegram style speech, nor did she omit function words. She did, however, change the syntax in such a way that it no longer corresponded to what could be expected in her native language. She used French grammar when speaking Dutch, but not when speaking English. Paragrammatic speech is generally fluent, and marked by complex sentences which contain function words, verbs (also finite ones), nouns, in short: all elements required for the construction of a well-formed sentence are present, but the speakers do not apply the grammatical rules as expected.

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5.5. SHORTCOMINGS AND LIMITATIONS

The results of this review should be interpreted with caution. The scarcity of comparable measures characterizing the case reports compelled us to limit the quantitative analysis of FAS. With a view to future diagnostics, it is hoped that linguistic manifestations, medical findings, medical history and psychiatric symptoms are documented in great detail, in order to enable a reliable FAS diagnosis and suitable therapeutic interventions.

5.6. CONCLUSION

This paper explored psychogenic FAS as a subtype of FAS. The following conclusions can be drawn: firstly, psychogenic FAS is related to the presence of a psychiatric or psychological disturbance in the absence of demonstrable neurological damage or an organic condition that might explain the accent. Secondly, psychogenic FAS occurs more in women than men, in an age range which is likely to be prone to depression and mental problems (25-49 years). Thirdly, psychogenic FAS is characterized by both suprasegmental and segmental changes. A deviant intonation (variable pitch) and a slow speech and articulation rate are the most typical prosodic features. At a segmental level, vowels are more affected than consonants. Future research should report on segmental and suprasegmental changes in as much detail as possible, in order to aid diagnosis based on semiological distinctions between neurogenic and psychogenic FAS. Fourthly, the remission of FAS seems to be related to resolution of comorbid positive psychiatric symptoms. Fifthly, psychodiagnostic testing – including symptom validity tests – is highly recommended with a view to suspected psychogenic FAS; not only in view of adequate therapy, but also for the interpretation of cognitive deficits, which may be aggravated as well. Sixthly, patients with psychogenic FAS often demonstrate linguistic features in speech and language that are not consistent with neurogenic speech/language disorders, e.g. in psychogenic cases, FAS can co-occur with a form of isolated 'pseudo-' agrammatism in unaffected fluent speech (different from agrammatism seen in non-fluent aphasic patients) and paragrammatism. Pre-FAS mutism has also been attested. Furthermore, language often shows code switching and language mixing which rarely occurs in polyglot aphasic patients.

Future research should work towards the validation of a set of criteria for psychogenic FAS via an extensive comparison with the neurogenic cognate. Moreover, in view of an efficient therapeutic guidance and clinical diagnosis, future research should focus on the treatment of non-organic *speech and language* disorders in large populations. We believe that a combination therapy focusing on the cognitive-behavioral problems on the one hand, and speech and language deficits on the other, may be beneficial in this population. The intricate symptomatology often gives proof of overlapping cognitive,

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psychological and speech problems, and the FAS is interpreted as an (indirect or direct) emanation of the underlying psychological disturbances.

5.7. ACKNOWLEDGMENTS

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CHAPTER 6

Perceptual accent rating and attribution in psychogenic FAS: some further evidence challenging Whitaker's operational definition[18]

[18] This study in this chapter was reported in Frontiers in Human Neuroscience: Keulen, S., Verhoeven, J., Bastiaanse, R., Mariën, P., Jonkers, R., Mavroudakis, N. & Paquier, P. (2016). Perceptual accent rating and attribution in psychogenic FAS: Some further evidence challenging Whitaker's operational definition. *Frontiers in Human Neuroscience*, *10*, 1-14.

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ABSTRACT

A 40-year-old, non-aphasic, right-handed, and polyglot (L1: French, L2: Dutch, L3: English) woman with a 12 year history of addiction to opiates and psychoactive substances, and clear psychiatric problems, presented with a foreign accent of sudden onset in L1. Speech evolved towards a mostly fluent output, despite a stutter-like behavior and a marked grammatical output disorder. The psychogenic etiology of the accent foreignness was construed based upon the patient's complex medical history, and psychodiagnostic, neuropsychological, and neurolinguistic assessments. The presence of a foreign accent was affirmed by a perceptual accent rating and attribution experiment.

It is argued that this patient provides additional evidence demonstrating the outdatedness of Whitaker's (1982) definition of Foreign Accent Syndrome, as only one of the four operational criteria was unequivocally applicable to our patient: her accent foreignness was not only recognized by her relatives and the medical staff, but also by a group of native French-speaking laymen. However, our patient defied the three remaining criteria, as central nervous system damage could not conclusively be demonstrated, psychodiagnostic assessment raised the hypothesis of a conversion disorder, and the patient was a polyglot whose newly gained accent was associated with a range of foreign languages, which exceeded the ones she spoke.

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6.1. INTRODUCTION

Foreign Accent Syndrome (FAS) is a speech-output disorder, which affects the segmental and suprasegmental characteristics of speech in such a way that a speaker is no longer able to make the correct phonetic/phonematic contrasts of his/her native language. The FAS speaker is interpreted by listeners to be a non-native speaker of his/her mother tongue, or – in some cases – as speaking a different dialectal variant. Numerous cases of FAS have been attested since Pierre Marie (1907) described the case of a Parisian man who started speaking with an Alsatian accent after having sustained an intracerebral hemorrhage, which Marie localized at the level of the left lentiform nucleus. Reeves & Norton (2001) were the first to explicitly link their schizophrenic patient's foreign accent (syndrome) to his psychotic exacerbations. Before them, Critchley (1964) and Gurd et al. (2001) had already hinted at a possible psychogenic etiology for the FAS their patients had developed. However, they did not label it as such, possibly due to a lack of objective proof, and because in the context of Whitaker's (1982) operational definition (**Table 6.1**), the possibility of a psychogenic FAS is excluded. According to Whitaker's criteria, indeed, FAS is strictly related to central nervous system damage.

Table 6.1. Whitaker's operational definition of FAS (Whitaker, 1982; pp. 196 and 198).

(1)	The accent is considered by the patient, by acquaintances and by the investigator, to sound foreign.
(2)	It is unlike the patient's native dialect before cerebral insult.
(3)	It is clearly related to central nervous system damage (as opposed to an hysteric reaction, if such exist).
(4)	And there is no evidence in the patient's background of being a speaker of a foreign language (i.e., this is not like cases of polyglot aphasia).

In 2005, Van Borsel et al. defended the hypothesis of a psychogenic FAS in their 32-year-old female patient, who presented with FAS, as well as with subtle grammatical anomalies. Medical history revealed she had suffered from depression and suicidal ideation. A neurological and radiological work-up did not reveal any neurological deficit. Other psychogenic case studies would follow (Verhoeven et al., 2005; Poulin et al., 2007; Reeves et al., 2007; Haley et al., 2010; Cottingham & Boone 2010, Jones et al., 2011; Lewis et al. 2012; Roy et al., 2012; Polak et al., 2013). Close inspection of the FAS case studies – irrespective of the etiological substrate – reveals that this disorder rarely occurs as a 'stand-alone phenomenon'. Rather, there is a rich spectrum of possible comorbid speech and language impairments that can accompany FAS. The most common comorbid speech and language disorders of neurogenic FAS are: dysarthria, apraxia of

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speech, and aphasia, mostly of the non-fluent type, although the fluent type has also been reported. In addition, muteness has been reported as a speech disorder frequently preceding FAS (**Table 6.2**). Furthermore, specific language impairment (SLI), developmental apraxia of speech (DAS), and agrammatism (mainly in a context of aphasia) have been noted.

Table 6.2. Overview of the comorbid speech and language disorders in neurogenic FAS cases

Comorbid speech and language disorders	References
Dysarthria	e.g. Monrad-Krohn 1947; Nielsen & McKeown, 1961, case 1 & 2; Whitty 1964; Schiff et al., 1983; Graff-Radford et al., 1986; Berthier et al., 1991, case 1 & 2; Scianna et al. 2000
Apraxia of speech	e.g. Whitty 1964; Ingram et al., 1992; Takayama et al., 1993, Laures-Gore et al., 2006, case 1
Aphasia: fluent	e.g. Kwon & Kim, 2007; Katz et al., 2008
Aphasia: non-fluent	e.g. Monrad-Krohn 1947; Nielsen & McKeown 1961, case 1; Graff-Radford et al., 1986; Ardila et al., 1988; Berthier et al., 1991, case 3 & 4
Mutism (pre-FAS)	e.g. Gurd et al., 1988; Berthier et al., 1991, case 1 & 2, Roth et al., 1997; Carbary et al., 2000; Munson & Heilman, 2005

In psychogenic FAS, the only comorbid speech/language impairments that have been attested over the years are a 'pre-FAS'-muteness (Van Borsel et al., 2005; Jones et al., 2011), and grammatical anomalies (Van Borsel et al., 2005; Verhoeven et al., 2005; Poulin et al. 2007; Cottingham & Boone, 2010). The previously mentioned 32-year-old right-handed patient reported by Van Borsel et al. (2005) presented with grammatical errors, not explicable by any neurological damage. Aberrant realizations concerned substitution errors (mainly affecting nouns and verbs), omissions (especially affecting auxiliaries, prepositions and articles), and a dyssyntaxis. Importantly, the authors note that this pattern of grammatical errors did not conform to the pattern typically seen in Broca (agrammatism) or Wernicke (paragrammatism) aphasia. FAS in their patient found its expression through segmental alterations (e.g. "devoicing of voiced consonants", "cluster reduction of r-clusters", "initial consonant deletion of /h/" ; p. 423) and suprasegmental alterations ("improper word stress", "improper sentence stress", "a tendency towards scanning speech"; p. 423). Two years later, Poulin et al. (2007) (see also: Roy et al., 2012) diagnosed FAS in a 74-year-old, bipolar patient. Although they doubted the psychogenic origin of FAS, consensus as to the psychogenicity in their patient was

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subsequently reached among other authors (see: Mariën et al., 2009; Haley et al., 2010; Jones et al., 2011; Lewis et al., 2012). Other instances of psychogenic FAS in bipolar patients would follow (Reeves et al., 2007; case 2). Poulin et al.'s (2007) patient demonstrated 'mild agrammatism'. In contrast to what is generally seen in Broca's aphasia patients with a marked agrammatism, the speech of their patient was fluently[19] produced, although it was telegraphic in structure. Function words as well as bound grammatical morphemes were omitted. Unfortunately, the possible occurrence of a (multimodal) grammatical disorder in written language production was not investigated. In 2010, Cottingham & Boone described a 36-year-old woman implicated in a motor vehicle accident (MVA), who developed an Eastern European-like accent three years after the MVA occurred. She too developed a 'telegraphic style' of speech. A combination of different arguments pleaded for the psychogenic etiology of her accent shift. First, there was the late onset of the accent (3 years post-MVA). In addition, there were no demonstrable anomalies on MRI and EEG. Furthermore, the patient exhibited a left-sided give-way weakness. Linguistically, she demonstrated difficulties in sentence repetition (10 days post-MVA), which were limited to the clinical test setting, as well as improbable error patterns (splitting numbers into separate digits)[20]. There were irregularities in the (aberrant) intonation pattern and inconsistencies in the grammatical disorder (deleting a preposition in one sentence, using it in the following sentence and then deleting it again). Lastly, her answers on the Minnesota Multiphasic Personality Inventory (MMPI-2) (Butcher et al., 1989), although less conclusively than expected (possibly influenced by a defensive stance), indicated a hysterical personality orientation (suggesting conversion disorder).

The current paper adds to the literature on psychogenic FAS by presenting a new case challenging Whitaker's (1982) operational definition. We report a 40-year-old, non-aphasic woman, with a twelve-year history of addiction to psycho-active substances, who presented at the Erasme University Hospital neurology department between 2010 and 2013, with a complex and diverse set of symptoms mainly perturbing her gait and language. Alterations affecting her oral verbal output were stutter-like behavior, (atypical) grammatical errors[21], as well as FAS. Based upon an analysis of her complex medical history, and the psychodiagnostic neuropsychological, and neurolinguistic assessments, we advance the hypothesis of a psychogenic etiology. This case report also demonstrates that identifying the provenance of the perceived accent foreignness depends on the listener's subjective impression.

[19] The American Speech-Language-Hearing Association (ASHA) defines fluency as "the aspect of speech production that refers to the continuity, smoothness, rate, and/or effort with which phonologic, lexical, morphologic, and/or syntactic language units are spoken" (American Speech-Language-Hearing Association, 1999).

[20] For instance, the number 11 was uttered as "one one".

[21] Although the grammatical disorder is a noteworthy aspect of the patient's language profile, the analysis of its multimodal characteristics is beyond the scope of the current study, and will be reported elsewhere.

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6.2. BACKGROUND

In a study on the pathophysiological mechanisms of different speech disorders, Whitaker (1982) assigned four characteristics to the speech disorder he coined 'Foreign Accent Syndrome' (**Table 6.1**). As is clear from our introduction, many case reports have defied one or more criteria proposed by Whitaker. This has been an incentive for the conceptualization of a distinctive taxonomic variant of FAS, *psychogenic FAS*, which is instigated by psychological or psychiatric problems (Verhoeven & Mariën, 2010a).

The aim of the current study is twofold: (1) based upon the medical history, the symptoms at presentation, the neurocognitive work-up, as well as the psychodiagnostic and neurolinguistic assessments, we argue that the nature and evolution of the patient's speech/language symptoms are highly indicative of a psychogenic etiology; (2) we experimentally corroborate the hypothesis of FAS by performing an accent rating and attribution task (Verhoeven et al., 2013).

6.2.1. Patient and medical history

The patient gave informed written consent to report her data according to the standards and regulations established by the ethics committee of the Erasme Hospital (ULB)[22].

The patient is a 40-year-old, right-handed, Belgian woman with 13 years of education. She is an unbalanced polyglot speaker: she was raised in French (L1) and Dutch (L2) as an early bilingual, and learned English (L3) at secondary school. French is her everyday language. She sustained a cerebral concussion after a fall at age 17. She had suffered from severe addiction to multiple opiates and psychoactive substances (cocaine, lsd, cannabis, …) for a period of twelve years (1988-2000). In 2003, she benefited from a last inpatient withdrawal treatment, after which she self-admittedly stated to have been clean. In 2005, she was admitted to the same psychiatric institution because of somatization, insomnia, anxiety, underfeeding, and aboulia, probably resulting from an anxio-depressive decompensation. She was considered to exhibit a histrionic personality disorder. An EEG was normal. She was discharged after a month of intensive psychotherapy, and remained under antidepressant and anxiolytic medication. She underwent surgery for a C5-C6 cervical hernia in 2008.

In February 2010, the patient was readmitted to the psychiatric institution because of speech problems of sudden onset[23], characterized by telegraphic speech, stuttering, and a change of accent especially when speaking French. She also complained of attention problems, nuchal pain, and arthralgia. She presented with non-rhythmic myoclonic jerks in lower limbs disturbing her gait, but clinical neurological examination revealed no motor or sensory deficits. Tendon reflexes were normal, and there was

[22] Regulations and procedures to be found at http://www.erasme.ulb.ac.be/page.asp?id=9536&langue=FR.

[23] These speech problems showed up when the patient was refused additional financial support by an official social insurance company.

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no cerebellar dysmetria. CT scan and MRI of the brain were normal, as was an EEG. Clinical biology tests revealed no abnormalities. Bone scintigraphy, cervical CT scan, and echography of uterus were all normal. Incidentally, at one occasion, she was noticed to speak normally during a temper tantrum caused by a feeling of not being taken seriously by the nursing staff.

In June 2010, the patient was seen at the neurological outpatient clinic for complaints concerning gait and speech. The gait and language abnormalities could not be explained by any neurologically induced deficits, and the hypothesis of a conversion disorder as well as Münchausen syndrome was formulated. In July 2010, the patient was hospitalized for largely the same complaints as the month before: (unstable) gait, backache, as well as impaired speech and language. The most striking speech symptoms consisted of a telegraphic output and stuttering (affecting her French), along with a change of accent (all formally attested during neurolinguistic investigation). Clinical neurological examination, CT scan of the brain, and clinical biology tests (HIV, mycoplasma, HCV, HBV, syphilis and Borrelia) were completely normal. An MRI of the brain, performed prior to the current admission, was reported to be normal except for a discrete cortico-subcortical atrophy. An EEG was inconclusive because of the presence of muscular artifacts. Because of the multiple complaints of cervical and joint pain, a second follow-up was initiated at the outpatient algologic clinic. In September 2010, she was initially seen at the neurological outpatient clinic, but was hospitalized because she repeatedly fell (admitted after a fall out of a wheelchair) and had diffuse pain complaints (especially situated near the cervical discs). Psychiatric complaints were noted after admission. The patient showed a behavioral regression limiting her autonomy.

In April 2011, the patient was again admitted for approximately one month to the psychiatric ward because of depression, insomnia, and regression of her physical state. When hospitalized, medical staff equally noted a behavioral regression to an infantile state: the patient was incontinent (wore diapers), had cuddly toys in her hospital bed, used a pacifier, and kept herself in fetal position. Clinical neurological examination, CT scan of the brain, and an EEG were all normal. In May 2011, she received a full neurolinguistic work-up (*see below*), which demonstrated deficits affecting all language faculties. The foreign accent, articulatory efforts, and stuttering had diminished compared to June 2010. The grammatical output disorder, that affected her (fluent) speech as well as writing, was still perceptible. The neurolinguist concluded that the speech and language symptomatology was unlikely to be caused by a neurological disorder. The follow-up notes of the algologist until February 2012 did not mention any improvement of speech and language.

In August 2012, she was seen at the neurological outpatient clinic. At that time, she was wheelchair-bound due to sudden immobility of the lower limbs and hypoesthesia of the left hemicorpus. A last neurolinguistic work-up was realized, which demonstrated that the grammatical disorder was still present in writing, but no longer in speech. The foreign accent also had disappeared, and stuttering had remarkably diminished compared to May 2011. Language problems had – according to the patient –

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spontaneously receded after she woke up from an appendectomy under general anesthesia performed one month earlier in a peripheral hospital.

The last time the patient was seen at the neurological outpatient clinic in July 2013, oral language production was normal. The patient presented with a complex clinical picture associating a fibromyalgic syndrome, osteo-articulatory pain, arthrosis, and a cervical discopathy. Because of the spontaneous resolution of her speech and oral language problems, the patient no longer sought neurological advice at our institution.

6.3. METHODS

6.3.1. Psychodiagnostic assessment

Psychodiagnostic assessment was conducted in 2010 by means of a structured interview, the Rorschach Test (Rorschach, 1921; Rorschach & Oberholzer, 1923) and the Object Relations Technique (Shaw, 2002). Results revealed passive self-reflection and infantile tendencies in thought, which had not (yet) found expression in her actions (this was the case in April 2011; see medical history). The psychodiagnostic examination did not indicate a psychological dissociation. According to her Rorschach test results, the patient had regressed to an 'archaic' stadium, which caused her to be nervous and which could have been incited by a fear to enter 'the adult world', possibly due to traumatic events she experienced as a child (tumultuous relationships with her parents and relatives). Based upon the neurological and psychiatric examinations, and given the numerous somatic complaints for which no organic lesions could be demonstrated, the patient was considered to suffer most likely from a 'hysterical conversion disorder', although this was not substantiated by formal psychodiagnostic testing (she refused to be administered the MMPI).

6.3.2. Neuropsychological assessment

Standardized neuropsychological tests were carried out in 2010 (**Table 6.3**). The patient had an estimated pre-morbid IQ of 92 (Beauregard, 1971), which corresponded to an IQ of 91 as measured by the Raven Progressive Matrices (Raven, 1996). Verbal reasoning was normal according to the WAIS-Similarities subtest (Wechsler, 1970). The patient's short-term memory was normal in the visuospatial modality as measured by the Corsi block-tapping test (Milner, 1971) and the Violon Beehive Test (Violon & Wijns, 1984), but slightly defective in the verbal modality according to the WAIS-Digit Span (Wechsler, 1970). Delayed memory was impaired both in the visuospatial modality as assessed by the Benton Visual Retention Test (Benton, 1953) and the Rey-Osterrieth Complex Figure Test (ROCF) (Rey, 1941), and in the verbal modality in agreement with the Rey Auditory-Verbal Learning Test (RAVLT) (Rey, 1964). Free verbal recall was nor-

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mal according to the Wechsler Memory Scale-Logical Memory (Wechsler, 1969), but the RAVLT (Rey, 1964) showed decreased verbal learning. Visuoconstructive skills were normal and there were no signs of visual neglect on the ROCF (Rey, 1941). The patient showed normal performance on Part A of the Trail Making Test (Reitan, 1992; Godefroy, 2008), but Part B indicated decreased speed for attention and sequencing. This could not be confirmed by the WAIS-Coding subtest (Wechsler, 1970). Of note, during the neuropsychological assessment, the psychologist also discerned a 'German/Slavic'-like accent, along with a severe grammatical anomaly in spontaneous speech.

Table 6.3. Neuropsychological test results (September 2010) (pc. = percentile)

TEST	Raw score (/max. score) (st.= standard score)	Percentile	Mean	SD
MEMORY				
Wechsler Memory Scale				
Verbal span (direct/reverse)	7		10.54	1.92
Logical memory	9		10.43	3.07
Block tapping test of Corsi	4		5.11	1.01
Benton drawings				
Immediate recall	3 (/10)			
Delayed recall	10 (/25)			
Rey Complex Figure				
Model	I	Pc. 50-100		
Time	2'	Pc. 100		
Score	14	Pc. < 10		
The beehive test (Violon)				
Memory				
1st trial	2		5.60	2.72
2nd trial	8		7.80	2.07
3rd trial	8		8.65	2.21
4th trial	10		9.10	2.05
5th trial	10		9.45	1.32
15 words of Rey				
Recall: total *n* words	42	Pc. <25		
1st trial	8	Pc. 50		
3rd trial	8	Pc. 0		
5th trial	6	Pc. 0		
ESTIMATED PREMORBID IQ				
Verbal automatisms of Beauregard	21(/40)	Pc. 25 (IQ: 92)		

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TEST	Raw score (/max. score) (st.= standard score)	Percentile	Mean	SD
INTELLECTUAL FUNCTIONS				
Raven matrices	30(/60) (IQ: 91) (time: 32')			
WAIS – Similarities	17(/26) (st.= 10/20)			
PRAXIS				
Rey Complex Figure				
Model	I	Pc. 50-100		
Time	2'10"	Pc. 75		
Score	26	Pc. <10		
CONCENTRATION, ATTENTION, MENTAL CONTROL				
WAIS coding	35 (st.= 8/20)		33.55	1.4
EXECUTIVE FUNCTIONS				
Trail Making Test				
Time (A)	35"	Pc. 50-75	31"	12
Error (A)	0	Pc. 5-75	0.12	0.45
Time (B)	135"	Pc. >95	66"	24
Error (B)	2	Pc. 5	0.14	0.46

6.3.3. Neurolinguistic assessment

Neurolinguistic assessments took place in July 2010, May 2011, and August 2012 by means of a series of standardized tests (**Table 6.4**), and repeatedly failed to evidence aphasia.

Auditory comprehension

Auditory comprehension was assessed using the French version of the Boston Diagnostic Aphasia Examination (BDAE) (Mazaux & Orgogozo, 1983) and the shortened Token Test (De Renzi & Faglioni, 1978). Except for the Token Test, results were well within the normal range on the three occasions. Both Token Tests administered (2010 and 2011) were slightly defective because of confusions between tokens in otherwise correctly executed commands.

Oral expression

Oral expression was assessed by means of the French version of the BDAE (Mazaux & Orgogozo, 1983) and the Bachy 36-items naming test (Bachy-Langedock, 1989). In July

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2010, performance on most oral language tasks was severely hampered by a complex speech disorder combining: (a) a stutter-like behavior with articulatory efforts in initiating words, associated with spectacular facial synkinesias, (b) an impressive grammatical disorder in spontaneous speech and across all tests administered (including oral repetition and reading aloud tasks) that was observed in L1 but not in L2 and L3, and (c) a foreign accent which was perceived by the neurolinguist as English or Slavic, and which similarly only affected her native language. Of note, the patient did not produce one single paraphasia in the sentences generated during the entire oral language assessment, and obtained normal results on automatized sequences (counting, days of the week, months of the year), a responsive naming task (word finding upon orally presented questions), a body-part naming task, and a semantic verbal fluency task (1 and 2 minute-generation of animal names).

Overall performance in oral language was roughly similar in May 2011, although the foreign accent, articulatory efforts, and stutter-like behaviors had considerably diminished at that time. However, a prominent and paradoxically fluently produced grammatical disorder was still noticed in spontaneous speech and during all language tasks. Again, paraphasic errors were not observed.

In August 2012, one month after an appendectomy under general anesthesia, the patient was referred to the neuropsychological department by her neurologist, who was astonished by the unexpected and unexplained improvement of her oral language skills. The grammatical disorder in spontaneous speech and oral language tasks had completely disappeared, as had the foreign accent. Sporadically, a discrete and short-lasting stuttering was observed. Results on oral language tasks were well within normal limits, except for a persistently weak performance on visual confrontation naming and a decreased generation of animal names (paradoxical reduction of semantic verbal fluency in association with a spectacular improvement of oral expression) (**Table 6.4**).

Reading

Reading aloud in 2010 and 2011 (assessed by means of the BDAE) was effortful mostly because of the stutter-like symptoms, and was characterized by a foreign accent. Moreover, reading sentences was contaminated by massive grammatical errors. The words composing the sentences, however, were correctly read. As was the case in spontaneous speech, in 2012 reading aloud had completely normalized. Reading comprehension of sentences and paragraphs was normal in 2010 and 2011. Unexpectedly, the patient performed worse at the time oral language and reading aloud had normalized (**Table 6.4**).

Writing

In written language production (assessed by means of the BDAE), graphomotor skills and writing words upon dictation were normal at the time of the three language evalu-

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ations. Writing sentences upon dictation was altered by grammatical errors (omissions of grammatical words, use of infinitive verbs), but the words themselves were written flawlessly. The written description of the Cookie Theft picture remained grammatically impaired over time, although, again, all individual words were spelled correctly.

Table 6.4. Neurolinguistic test results.

TEST (/max. score)	SCORES July 2010	SCORES May 2011	SCORES August 2012	CUTT-OFF
Oral comprehension				
Word discrimination BDAE (/72)	70	68.5	71	67
Body part identification BDAE (/20)	18	18	18	18
Commands BDAE (/15)	14	15	15	13
Token Test (/36)	27	27	31	29
Oral expression				
Non verbal agility BDAE (/12)	6	NA	7	9
Verbal agility BDAE (/14)	6	6	11	11
Automatized sequences BDAE (/8)	8	6	NA	6
Repetition BDAE				
- Words (/20)	15	18	20	18
- High probability repetition (/8)	3	3	8	6
- Low probability repetition (/8)	2	2	8	6
Responsive naming BDAE (/30)	29	30	30	27
Naming Bachy 36 items (/36)	27	31	29	35
Body part naming BDAE (/30)	30	27	30	24
Verbal Fluency (animals) (1 min.)	18 (pc. 25)	15 (<pc. 10)	11(<pc. 10)	16.5
(2 min.)	26	22	17	24.5
Reading				
Word reading BDAE (/30)	27	24	30	30
Sentence reading BDAE (/10)	1	4	10	8
Symbol and Word Discrimination BDAE (/10)	NA	8	NA	10
Word recognition BDAE (/8)	NA	8	NA	6
Comprehension of oral spelling BDAE (/8)	NA	8	NA	6
Word/picture matching BDAE (/10)	10	NA	NA	10
Reading sentences and paragraphs BDAE (/10)	9	8	6	7
Writing				
Writing Mechanics BDAE (/5)	5	5	5	5
Serial writing BDAE (/47)	NA	46	NA	47
Primer-level dictation BDAE (/15)	NA	15	15	15
Spelling to dictation BDAE (/10)	8	8	7	6
Sentences to dictation BDAE (/12)	NA	NA	10	10
Written confrontation naming BDAE (/10)	8	10	NA	7
Narrative writing BDAE (/5)	3	3	3	4

NA= Not Administered, Pc.= percentile

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6.3.4. Phonetic assessment

The first author (SK) performed a perceptual analysis of five minutes of spontaneous speech during which the patient explained her medical history, in order to seek which segmental and suprasegmental features could have induced or at least reinforced the impression of accent foreignness. To this purpose, the excerpt was transcribed into International Phonetic Alphabet. As the patient's foreign accent was judged to have diminished as of 2011, a sample was selected from the recordings made in 2010.

Perceptually, the patient appeared to realize the French uvular /R/ as an English diphthong. For instance, the verb *faire* (/fɛR/) (to do) was pronounced as /feəʳ/. On other occasions, she used excessive alveolar trill (as, for instance, in Italian, Spanish, or Russian) instead of uvular rhoticity. Other segmental errors consisted of additions of [r] (devoir→devroir) and schwa (plus→p*ə*lus) (epenthesis). The patient sometimes used a voiced velar fricative (/ɣ/) instead of the voiced velar plosive /g/ (e.g., /ɣrɑm/ for /*g*Rɑm/ or 'gram' in English), which could have induced the impression of a Dutch/Flemish-like accent. Moreover, she produced voiceless and voiced ejective consonants as, for instance, in /k'ɒm/ (*comme*; like), /bRɛIk'dæns/ (*breakdance*), /beg'ɛje/ (*bégayer*; to stutter). Ejectives are highly uncommon in European languages, and occur in some languages in the region of the Caucasus and the Americas (Hayward, 2013). The patient equally spoke with a strangled voice, probably reinforced by the repeatedly produced egressive, glottalic airflow which caused the realization of the ejectives, instead of the typical, expected pulmonic egressive airstream. Intonation of speech was aberrant. Word accent was sometimes wrongfully placed (e.g., *beau*coup; many). Melody of speech was equally altered in 2010, and there were sudden excursions of speech intensity.

6.4. EXPERIMENT

6.4.1. Aim

A perceptual accent rating and attribution experiment was set up with the purpose of disclosing (a) whether a group of French-speaking listeners judged the patient to speak with a foreign accent, (b) which accents could possibly be identified in the FAS speaker's speech, and additionally (c) how native and non-native speakers of French could be identified. Because of the severe speech impediment suffered by the patient, we decided to apply Dankovičová & Hunt's (2011) procedure to select the stimuli (*see below*).

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6.4.2. Methods

6.4.2.1. Materials and samples

This study consisted of a perceptual experiment in which 25 French-speaking students in French linguistics at a francophone university in Brussels – who were not formally acquainted with speech pathologies of any kind – blindly assessed the (foreign) accent and linguistic background of six speakers. One speaker was the FAS patient, her stimuli were mixed with stimuli from five other speakers: one was a native French-speaking Belgian woman stemming from the same geographic area as the patient, and four others were non-native speakers of French with an audible foreign accent.

The selected stimuli were retrieved from a recorded informal interview, which took place in 2010 in the context of neurolinguistic testing. The patient explained her medical history, symptoms, and the chronology of events. Nine isolated words and six grammatically correct utterances were selected and edited as to ensure full anonymity (see also: Dankovičová & Hunt, 2011). Only correct utterances were chosen in order to avoid any possible artifacts in the listeners' judgments. In total, ninety stimuli were presented to the raters (15 stimuli x 6 speakers). Files were adjusted for the purpose of assessment using PRAAT, version 5.4 (PRAAT for Mac; Boersma & Weenink, 2014).

6.4.2.2. Control speakers

Five female control speakers (**Table 6.5**) read the words and utterances selected from the patient's interview. Recordings were made with a Marantz Professional PMD 661 portable recorder, and adjusted via PRAAT (Boersma & Weenink, 2014). The non-native speakers of French were respectively of Belgian (Dutch), English, German and Chinese origin. In accordance with Verhoeven et al.'s (2013) methodology, their foreign accents had not been matched to those the medical staff had tentatively reported in the patient. It was assumed that most listeners would be acquainted with the control speakers' accents.

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Table 6.5. Demographic data of speakers (FAS and controls) in the perceptual accent rating experiment, including an indication of the level of French, CEFR= Common European Framework of Reference for Languages (Council of Europe, 2001).

Nature	Gender	Age	Country of Birth	Mother tongue	Level in French (CEFR)
FAS	F	40	Belgium	French	_
Control 1	F	37	Belgium	French	_
Control 2	F	48	Belgium	Dutch	B1+
Control 3	F	52	Germany	German	B1+
Control 4	F	48	China	Mandarin Chinese	A2+ / B1
Control 5	F	42	United Kingdom	RP English	A2+ / B1

6.4.2.3 Stimuli and assessment

Total sample time was 25 min. and 26 sec. The stimuli were separated from one another by a 15 second interval to allow for judgment. The sample consisted of 15 'blocks' in which each stimulus was uttered by all six speakers in pseudo-random order. Stimuli were presented only once, so each speaker recurred 15 times.

Before hearing the speech samples in open field at their institution, the listeners received the test instructions, and completed demographic information about themselves (age, gender, country of birth, time living in Belgium if not born here, mother tongue, and other spoken languages including an indication of proficiency in these languages) on a questionnaire. They were asked to rate the speakers' degree of French-speaking 'nativeness' on a seven-point scale: 1 = 'definitely *not* a native speaker of French'; 7 = 'definitely a native speaker of French'. In case the rating was < 7, listeners were asked to identify the speaker's mother tongue.

6.5 RESULTS

6.5.1 Demographic results

Among the 25 raters (16-25 years old; mean age: 19 years and 3 months; 11 males and 14 females), one participant was born in England, two in Luxemburg, and one in Mali. However, they all were raised and educated in French, except for the English student (aged 17), who was raised bilingually (French - English) but had been living in the French-speaking part of Belgium for 16 years.

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6.5.2 Accent rating results

Results were loaded into SPSS version 22 for Mac OS X (IBM Corp., 2013). First, inter-rater reliability was calculated for each speaker. As we had 25 different raters, this was examined by means of an intra-class correlation coefficient (ICC). As each item was assessed by each rater, and raters were randomly selected (sample selection, not population), the two-way random model was applied, checking for agreement implying that systematic differences between raters were taken into account. Results demonstrated that for FAS ICC(2,25)=0.77, for French ICC(2,25)=0.798, for Dutch ICC (2,25)=0.948, for German ICC(2,25)=0.936, for Chinese ICC(2,25)=0.936, and for English ICC (2,25)=0.713. These are acceptable values.

Mean scores, medians, standard deviations, minima, maxima, ranges and interquartile ranges are provided in **Table 6.6.** Based upon descriptive statistics, the French-speaking control appeared to be strongly associated with one extreme end of the continuum ($\bar{x}$=6.653, σ =1.043, M=7; score 7 = 'definitely a native speaker of French'), whereas the English-speaking control was clearly situated at the opposite extreme ($\bar{x}$=2.056, σ =1.589, M=1; score 1 = 'definitely *not* a native speaker of French'). The FAS patient, too, was associated more often with an elevated degree of foreignness ($\bar{x}$=2.288, σ =2.166, M=1). The remaining speakers were situated in between, they apparently were the most difficult to qualify as they were equally associated with the greatest standard deviations (Dutch: $\bar{x}$=3.949, σ=2.451, M=4; German: $\bar{x}$=3.880, σ =2.422, M=3; Chinese: $\bar{x}$=3.136, σ =2.164, M=3).

Table 6.6. Perceptual accent rating experiment: mean score, median, standard deviation (SD), minimum (Min), maximum (Max), range and interquartile range for the patient and each of the control speakers.

Speaker	Mean	Median	SD	Min	Max	Range	Interquartile range
FAS	2.288	1.000	2.166	1.000	7.000	6.000	2.000
French	6.653	7.000	1.043	1.000	7.000	6.000	0.000
Dutch	3.700	4.000	2.452	1.000	7.000	6.000	6.000
German	3.880	3.000	2.422	1.000	7.000	6.000	6.000
Chinese	3.136	3.000	2.164	1.000	7.000	6.000	4.000
English	2.056	1.000	1.589	1.000	7.000	6.000	2.000

As a Kolmogorov-Smirnov test of normality showed that data were not normally distributed (for all speakers: $p<0.01$), non-parametric statistics were applied. A Kruskall-Wallis H test showed that there was a statistically significant difference among ratings

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for the different speakers (inter-speaker difference: H(5)= 778.751, $p < 0.000$). Further analysis (Mann-Whitney *U*-tests) was necessary to establish inter-speaker comparisons. There was a significant difference among all speaker ratings (**Table 6.7**), except in the case of the ratings for the FAS patient (M=1) versus the English-speaking control (M=1): *U*= 68166.50, *p*= 0.407, and ratings for the Dutch- (M=4) versus the German-speaking controls (M=4): *U*= 69469.500, *p*= 0.771 and as such: $p > 0.0033$ (corrected *p*-value, Bonferroni correction).

Table 6.7. Perceptual accent rating experiment: Mann-Whitney-*U* scores for the individual inter-speaker comparisons.

GROUP COMPARISON	*N*	MEAN RANK	SUM OF RANKS	MANN WHITNEY *U*	WILCOXON *W*	*Z*	*p*
FAS	375	222.03	83260.50	12760.500	83260.500	-21.146	0.000
FRENCH	375	528.97	198364.50				
FAS	375	298.54	111951.50	41451.500	111951.500	-10.301	0.000
DUTCH	375	452.46	169673.50				
FAS	375	300.17	112564.50	42064.500	112564.500	-10.093	0.000
GERMAN	375	450.83	169060.50				
FAS	375	324.95	121855.50	51355.500	121855.500	-6.879	0.000
CHINESE	375	426.05	159769.50				
FAS	375	369.78	138666.50	68166.50	138666.500	- 0.829	**0.407**
ENGLISH	375	381.22	142958.50				
FRENCH	375	490.93	184098.00	27027.000	97527.00	-16.233	0.000
DUTCH	375	265,05	97527.00				
FRENCH	375	493.14	184928.50	26196.500	96696.500	-16.506	0.000
GERMAN	375	257.86	96696.50				
FRENCH	375	525.89	197207.00	13918.000	84418.00	-20.324	0.000
CHINESE	375	225.11	84418.00				
FRENCH	375	550.17	206314.50	4810.500	75310.500	-23.428	0.000
ENGLISH	375	200.83	75310.50				
DUTCH	375	377.75	141655.50	69469.500	139969.500	-0.291	**0.771**
GERMAN	375	373.25	139969.50				
DUTCH	375	411.27	154225.00	56900.000	127400.00	-4.626	0.000
CHINESE	375	339.73	127400.00				
DUTCH	375	459.42	172281.50	38843.500	109343.500	-11.074	0.000
ENGLISH	375	291.58	109343.50				
GERMAN	375	408.90	153337.50	37787.500	128287.500	- 4.322	0.000
CHINESE	375	342.10	128287.50				
GERMAN	375	457.61	171605.50	39519.500	110019.500	-10.847	0.000
ENGLISH	375	293.39	110019.50				
CHINESE	375	429.47	161053.00	50072.00	120572.00	-7.233	0.000
ENGLISH	375	321.53	120572.00				

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A correspondence analysis[24] (Clausen, 1998) in which the FAS patient and the control speakers represent the first categorical variable, and the ratings attributed to them (7 = 'definitely a native speaker of French'; 1 = 'definitely *not* a native speaker of French') the second categorical variable, confirmed that the FAS speaker and the English-speaking control were more strongly associated with accent foreignness than the other non-native speakers of French (**Figure 6.1** and **Table 6.8**). The native French-speaking control was most strongly associated with French-speaking 'nativeness' (Figure 6.1).

Table 6.8. Perceptual accent rating experiment: correspondence table presenting the frequency of each response (either 1,2,3,4,5,6, or 7) for the patient and each of the control speakers. The data are transformed to vectors in a two-dimensional space (Figure 6.1).

CORRESPONDENCE TABLE							
Rating	Speaker						
	FAS	FRENCH	DUTCH	GERMAN	CHINESE	ENGLISH	Active Margin
1	246	4	97	100	136	217	800
2	30	5	52	46	47	53	233
3	21	5	37	43	52	45	203
4	10	2	24	24	31	15	106
5	11	15	28	39	36	28	157
6	5	25	26	15	25	6	102
7	52	319	111	108	48	11	649
Active Margin	375	375	375	375	375	375	2250

[24] Correspondence analysis is a technique, which evaluates "the association between two or more categorical variables by representing the categories of the variables as points in a low-dimensional space. Categories with similar distributions are represented as points that are close in the space, and categories that have very dissimilar distributions are positioned far apart" (Clausen, 1998: p. 2).

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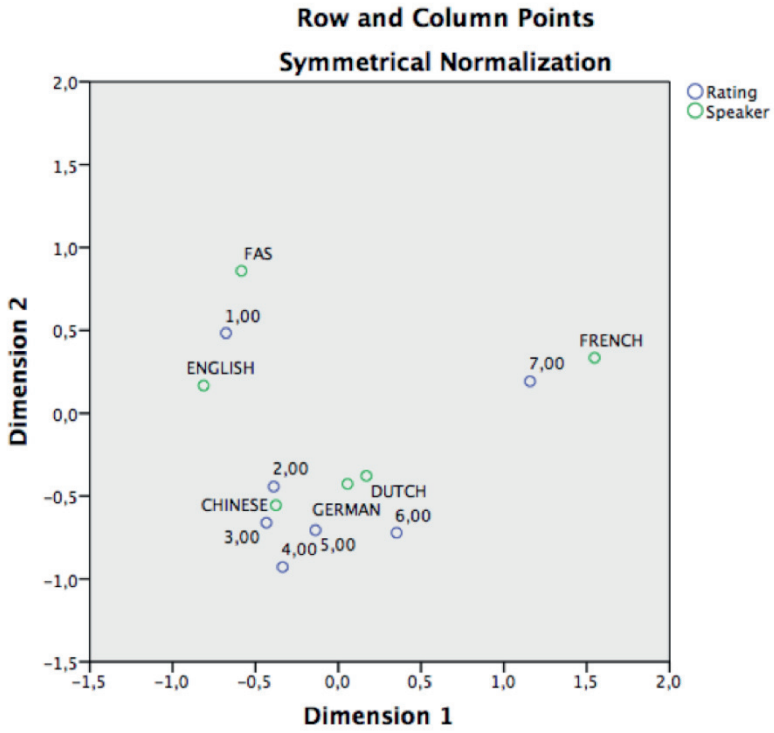
Figure 6.1. Perceptual accent rating experiment: correspondence analysis graphically displaying the accent dispersion and associated accent ratings in a two-dimensional space. The points represent a vector transformation of the data displayed in Table 6.8. The blue circles represent the accent rating, the green circles represent the speakers. Ratings were defined as column points, speakers as row points. The distances between the scores and speakers represent the strength of association between both values. Both FAS and English are more closely associated with 'Definitely non-native speakers of French' (= rating 1). French is (correctly) associated with 'definitely a native speaker of French' (= rating 7).



6.5.3. Accent attribution results

13/25 (52%) raters tried to identify the origin of the accent in those control speakers they judged not to be a native speaker of French (score <7 on the rating scale) (**Table 6.9**). **Figure 6.2** graphically displays the accent attribution of the 13 raters for all 15 stimuli per speaker (195 stimulus judgments per speaker). The native French-speaking control was recognized as a true native speaker of French in 185/195 (95%) of stimuli, whereas the FAS patient, who was also a native speaker of French, was perceived as such in only 46/195 (24%) of stimuli. In the native Dutch-speaking control, the difference between an association with a presumed French-like accent (*n*= 67/195, 34%) and a

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Dutch accent (*n*= 71/195, 36%) was minimal. The German-speaking control was identified as a native German speaker in only 20/195 (10%) of stimuli, whereas in 67/195 (34%) of stimuli she was considered a French speaker and in 59/195 (30%) of stimuli a Dutch speaker. The Chinese speaking control was identified as such in only 2/195 (1%) of stimuli. She was regarded as a native speaker of French in 34/195 (17%) of stimuli, and as a native speaker of Dutch in 55/195 (28%) of stimuli. Finally, the English-speaking control was properly identified as a native English speaker in 83/195 (43%) of stimuli, but was perceived as a Dutch speaker in 33/195 (17%) of stimuli. Speakers who were the least often associated with their native language (Dutch, German, Chinese) were mostly given scores of 3 or 4 on the rating scale, and had the greatest standard deviations.

Table 6.9. Perceptual accent attribution experiment: number of different accent origins associated with the patient and each control speaker.

	FAS	FRENCH	DUTCH	GERMAN	CHINESE	ENGLISH
Dutch	33	2	71	59	55	33
Luxembourgish	0	0	1	0	2	0
German	1	1	11	20	15	18
English	9	2	1	11	20	83
Norwegian	0	0	0	0	0	1
Danish	0	0	0	1	0	0
French	46	185	67	67	34	6
Spanish	18	1	13	8	19	10
Italian	3	0	10	2	12	9
Portuguese	1	0	1	3	3	8
Romanian	19	0	0	1	1	1
Greek	1	0	0	0	0	0
Russian	34	0	1	9	16	8
Macedonian	0	0	1	0	0	0
Polish	5	0	2	7	7	1
Turkish	0	0	0	0	1	0
Chinese	0	0	5	1	2	4
Japanese	1	1	0	0	1	8
Asian-sounding	0	0	0	0	1	2
Arabic	13	0	2	0	0	0
Unidentifiable stimuli	11	3	9	6	6	3
TOTAL (15 stimuli/speaker x 13 raters)	**195**	**195**	**195**	**195**	**195**	**195**

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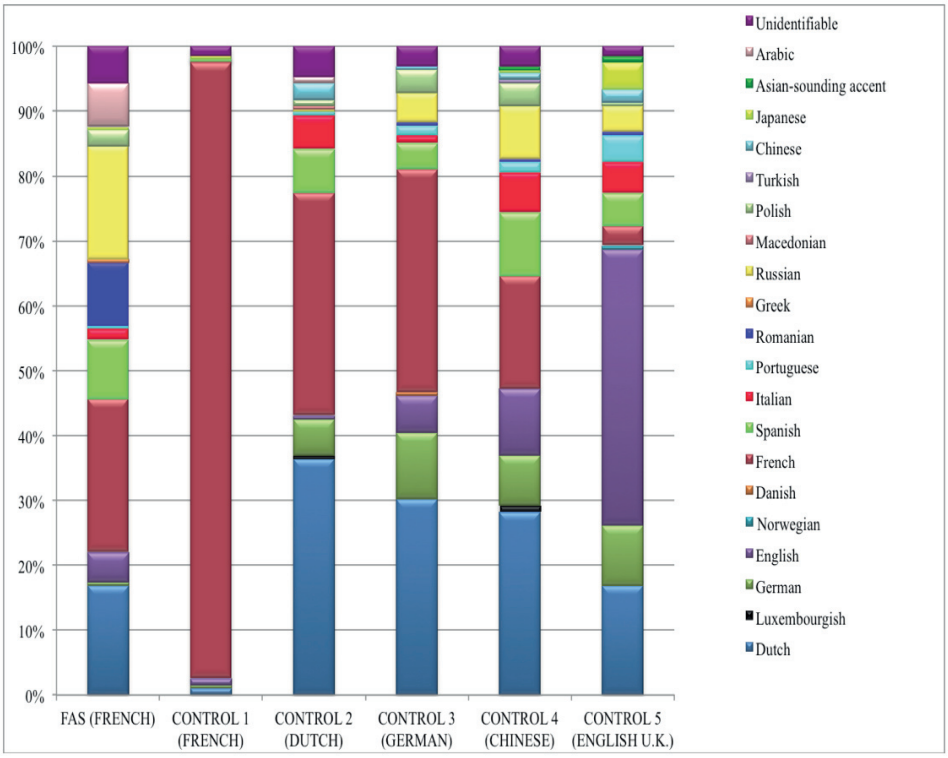
Figure 6.2 Graphic representation of the stratification of the different native languages which 13 native French speaking raters associated with the stimuli for the FAS patient and each of the control speakers. The FAS speaker was associated with 14 different accents including French. However, a comparison of the FAS patient with the native French speaking control clearly demonstrates that the raters identified the control's accent as their own in 95% of the stimuli vs. a mere 24% for the FAS patient (see paragraph 6.5.3).



Assumptions about the native language of all six speakers were the least stratified in the French-speaking control. The stratification of the number of putative native accents perceived in the other speakers was fairly similar. Both the FAS patient and the Dutch-speaking control were associated with 13 different languages. The English-speaking control's utterances were associated with 14 different languages, and those of the German-speaking participant with a total of 12 different languages. Accent attribution was most stratified in the Chinese speaker, her utterances being associated with no less than 15 possible languages (see **Figure 6.2**).

A majority of the raters surmised the FAS patient's utterances were produced by a person with a native Romance language: 87/195 (45%) of stimulus judgments were

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divided into 46 French, 19 Romanian, 18 Spanish, 3 Italian, 1 Portuguese. It should be noted, however, that the Romance language family was the most familiar to the raters, who were all students in French linguistics. When accent attributions to Germanic languages (43/195, 22%) and Slavic languages (39/195, 20%) were taken together – the language families the neurolinguist associated with the FAS patient – the difference between associations with Romance languages on the one hand and Germanic or Slavic languages on the other hand appears quite small.

6.6. DISCUSSION

The patient we report presented with a complex set of symptoms mainly affecting her gait and verbal output, and which could reasonably not be explained by any neurologically induced deficits. The most striking speech/language symptoms consisted of a stutter-like behavior, a grammatical disorder, and a change of accent in the absence of aphasia. Interestingly, these speech/language anomalies particularly altered her native language (L1: French) and were hardly observed in L2 (Dutch) and L3 (English). Two years after the initial neurolinguistic assessment, the oral speech/language deficits unexpectedly disappeared right after the patient woke up from general anesthesia induced for an appendectomy. While acknowledging the peculiar interest of the co-occurrence of accent foreignness and grammatical anomalies in psychologically induced speech-output disorders (Poulin et al., 2007; Van Borsel et al., 2005; Verhoeven et al., 2005), in the present study we purposely focused on the patient's change of accent (*see footnote 22*).

When a change of accent inducing an impression of accent foreignness originates from a pathological condition, it is called *Foreign Accent Syndrome*. FAS is "a motor speech disorder in which patients develop a speech accent which is notably different from their premorbid habitual accent" (Verhoeven & Mariën, 2010a; p. 600). Verhoeven & Mariën (2010a) classified FAS into three distinct taxonomical types: neurogenic, psychogenic, and mixed. In *neurogenic FAS*, the change of accent is associated with damage to the central nervous system. As such, it corresponds to the prototypical FAS as defined by Whitaker (1982). In *psychogenic FAS*, there is no evidence of neurological damage, and the accent change is ingrained in underlying psychological issues or psychiatric disorders. In *mixed FAS*, a neurologically induced accent change brings about psychological adjustments aiming at improving the authenticity of the newly acquired accent in order to create a more coherent new personality. This taxonomic differentiation has important implications for the management of treatment strategies.

In the current study, the patient's accent foreignness was affirmed by 25 independent, native speakers of French on the basis of an accent rating and attribution experiment. A majority of stimuli (76%) spoken by the FAS patient were assigned a non-French accent (whereas 95% of stimuli spoken by the French-speaking control were allocated a French accent). The stratification of the number of putative accents perceived in the FAS patient and the non-French-speaking controls was fairly similar (**Figure 6.2**). This

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finding also demonstrates the patient's strong accent foreignness, and corroborates the results of the phonetic assessment, which identified several segmental and suprasegmental transformations affecting the patient's speech output.

We consider the patient reported in the present study to represent an instance of psychogenic FAS for several, not mutually exclusive reasons:

1. The sudden and unexpected remission of all oral verbal output anomalies immediately after waking up from general anesthesia seems hard to explain on neurological grounds. Although the impact of general anesthesia on cognitive functions is still a matter of opinion (Guay, 2011), one would expect such an impact, if any, to induce (transitory) post-operative cognitive defects rather than improvements (Monk et al., 2008).

2. The selective, post-operative normalization of oral expression compared to the persistent grammatical disorder in written expression remains puzzling, especially in the absence of aphasia. Typically, spoken and written productions display similar error patterns in agrammatic aphasia subjects (Goodglass, 1993; Turkstra & Thompson, 2011). To the best of our knowledge, the exceptional instances in which a production deficit selectively spared grammatical utterances in an output modality while selectively affecting them in the other, have only been reported in the context of aphasia (Miceli et al., 1983; Goodglass, 1993; Miceli, 1999; Rapp & Caramazza, 1997; Vandenborre & Mariën, 2014).

3. In the absence of aphasia, the presence of an accent change, a grammatical disorder, and a stutter-like behavior selectively disrupting the patient's mother tongue while preserving L2 and L3 is also remarkable. Linguistic deficits disturbing one language more than the other(s) in multilingual speakers are a well-known phenomenon in the context of bilingual or polyglot aphasia (Paradis, 1995; Fabbro, 2001, 2002; Leemann et al., 2007; Gatignol et al., 2009). Up to the present, however, it remains speculative whether such selective speech output defects are also to be expected in non-aphasic, brain-lesioned patients.

4. Although the analysis of the patient's grammatical disturbances falls outside the scope of the current study, the bizarre and inconsonant pattern of her speech errors might shed an additional light on the nature of her verbal output disorder. As was the case in Cottingham & Boone's (2010) patient, the current patient also inconsistently split numbers into separate digits. For instance, she admitted having been addicted to psycho-active substances for 12 years (pronounced "douze"; *twelve*), which caused her to lose weight and to weigh 39 kilos (pronounced "trois neuf" [*three nine*] instead of "trente-neuf" [*thirty-nine*]). When asked in what year she had been admitted to one of the psychiatric institutions, she answered "en deux zéro zéro un" (*in two zero zero one*) instead of "en deux mille un" (*in two thousand and one*).

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5. The patient's mood apparently also influenced the characteristics of her spoken utterances, as she was able to shout grammatically correct sentences without foreign accent or stuttering during a violent fit of anger. Short utterances of normally produced spoken language can be observed in aphasic patients displaying automatic-voluntary dissociations between automatic and prepositional language use (Basso, 2003). However, the patient's outcries in this context clearly do not match short and automated or highly learned utterances.

6. Lesion-induced language and communication impairments are known to be at risk of occasioning emotional, behavioral, and psychosocial problems (Carota et al., 2002). Miller et al. (2011) showed that in neurogenic FAS, the accent change impacted on the patients' daily functioning by generating feelings of loneliness, depression, frustration, and loss of confidence. In spite of the nature and severity of her expressive difficulties, however, the current patient never showed frustration or dislike in relation to her verbal output impairment. She enjoyed the attention her speech disorder received, and always willingly participated in the neurolinguistic assessments. She did not try to avoid social contacts.

7. Repeat neurological examinations could not demonstrate any neurological deficits, and structural brain imaging studies repeatedly failed to disclose supratentorial and infratentorial lesions. Unfortunately, we could not obtain functional brain imaging data in the patient. In the event of a metabolic dysfunction or other subtle lesions not detected by structural brain imaging, such investigations might possibly have revealed functional abnormalities (e.g., brain perfusion or metabolism defects) in morphologically undamaged regions of the motor speech production circuitry at the time the verbal output disorder was present (Moreno-Torres et al., 2013). However, we find it difficult to relate the putative occurrence of such unexplored functional abnormalities in the distributed anatomical network controlling motor speech production to the selective impairment of phonetic/phonological components perturbing only one of the three languages spoken by the patient. Alario et al. (2010) admittedly showed that cognitive-based syllabic representations are separate in early bilinguals but are shared across languages in late bilinguals. Early bilinguals would have independent cognitive syllable representations allowing them to approach separate monolingual phonetic patterns in L1 and L2 (this might possibly theoretically explain the foreign-accent selectiveness in our patient on cognitive grounds), and late bilinguals would use the same cognitive representations when speaking either language. Alario et al. (2010) assumed that cognitive syllable representation would originate from the bilingual speaker's earlier L1 experience, which would be appropriate for L1 but only approximate for L2. However, according to this hypothesis, if one accepts the possibility of foreign-accent selectiveness in L1, one would expect a similar accent-foreignness in our patient's L1 and L3 as she was a late learner of English. We could not observe the latter accent pattern in our patient.

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8. A possible lesion-induced disruption of neurotransmitter activity and a potential influence of pharmacological interventions have been hypothesized to play an imaginable role in the appearance and resolution of FAS symptoms (Moreno-Torres et al., 2013). These assumptions are grounded in the observations that on the one hand, increasing cholinergic activity has been shown to facilitate recovery from post-stroke aphasia and apraxia of speech (Berthier & Pulvermüller, 2011), and that on the other hand, discontinuing neuroleptics in schizophrenic and bipolar patients has been shown to engender psychotic exacerbations with recurrence of a co-occurring FAS (Reeves et al., 2007), whereas restoring dopamine antagonists resulted in rapid suppression of symptoms (Reeves & Norton, 2001). However, the patient we report was not treated with neuroleptics during follow-up at our institution, as she never displayed psychotic signs since she was seen for the first time in 2010. Two months before the appendectomy performed in a peripheral hospital, a treatment with amisulpride was initiated by her psychiatrist because of the emergence of a depressive mood. Yet there appeared to be no pharmacological effect on the speech output disorder, as this did not change characteristics, and spontaneously and dramatically receded only two months later, right after the surgical intervention.

Given the above-listed arguments, we strongly believe the accent foreignness in the reported patient to be of psychogenic origin. Although a conversion disorder could not formally be confirmed by means of an MMPI, repeat neurological and psychiatric observations and follow-ups all clearly pointed to psychogenic behavioral and speech/language disturbances.

6.7. CONCLUSION

In 1982, Whitaker proposed four criteria a patient should meet in order to be diagnosed with FAS (**Table 6.1**). In the current paper, we report on a non-aphasic patient with FAS who only partly satisfied these criteria. The patient's accent was – in accordance with Whitaker's first criterion – perceived as 'foreign' by medical staff, friends and relatives, as well as by a group of 25 independent, native French-speaking listeners who rated her accent in a perceptual rating and attribution experiment. However, the two following criteria were challenged, in that we could not find any evidence of a *clear* cerebral insult to explain the sudden arousal of the accent. In addition, the patient was an unbalanced polyglot speaker of three languages, which is defying Whitaker's fourth criterion. As regards this last criterion, in the current experiment the patient's accent was associated with no less than 13 different languages, indicating that in polyglot FAS patients, listeners do not necessarily attribute the provenance of the perceived accent to one of the languages spoken by the patient. In fact, identifying the origin of the perceived foreign accent in FAS patients appears to depend on the degree of exposure of listeners to foreign accents (Di Dio et al., 2006; Miller et al., 2006; Verhoeven et al., 2013).

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In the present study, it is also remarkable that the accent foreignness, along with the grammatical disorder and the stutter-like behaviors particularly affected the patient's mother tongue (French) whereas these anomalies were hardly observed in L2 (Dutch) and L3 (English). Furthermore, the occasional loss of foreign accent (and other speech anomalies) when the patient was emotionally distressed was quite noteworthy. In addition, the sudden and unexpected resolution of the foreign accent after the surgical intervention remained quite puzzling, as was the modality-specific recovery from the oral grammatical disorder (whilst written expression remained grammatically altered). These and other behavioral observations in the patient all pointed to a psychogenic disorder that further contests Whitaker's third criterion. The latter was also called in question by at least five reports of FAS in association with a conversion disorder published between 2005 and 2011 (Verhoeven et al., 2005, Tsuruga et al., 2008, Haley et al., 2010, Cottingham & Boone, 2010; Jones et al., 2011).

As we conclusively demonstrated, the patient reported here suffered from a speech output disorder which listeners perceived as foreign-accented. The origin of the patient's accent foreignness and her multilingualism led us to conclude that Whitaker's operational definition of what he called "Foreign Accent Syndrome" (Whitaker, 1982; p. 195) is too restrictive and outdated. Whitaker's criteria appear not to offer enough space to include the currently accepted taxonomic variants of FAS (Verhoeven & Mariën, 2010a). Even for the neurogenic subtype, the last criterion seems barely maintainable, as polyglot, brain-injured FAS patients have also been reported (Schiff et al., 1983; Avila et al., 2004; Paquier & Assal, 2007; Levy et al., 2011). These findings underscore the necessity for a solid clinical diagnosis in the light of further treatment.

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CHAPTER 7

Psychogenic Foreign Accent Syndrome: a new case[25]

[25] The study reported in this chapter was published in Frontiers in Human Neuroscience: Keulen, S., Verhoeven,J., De Page, L., Jonkers, R., Bastiaanse, R., & Mariën, P. (2016) Psychogenic foreign accent syndrome: A new case. *Frontiers in Human Neuroscience, 10*, 1-13.

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ABSTRACT

This paper presents the case of a 33-year-old, right-handed, French-speaking Belgian lady who was involved in a car accident as a pedestrian. Six months after the incident she developed a German/Flemish-like accent. The patient's medical history, the onset of the FAS and the possible psychological causes of the change in accent are analyzed. Relevant neuropsychological, neurolinguistic and psychodiagnostic test results are presented and discussed. The psychodiagnostic interview and testing will receive ample attention, because these have been underreported in previous FAS case reports. Furthermore, an accent rating experiment was carried out in order to assess the foreign quality of the patient's speech. Pre- and post-morbid spontaneous speech samples were analyzed phonetically to identify the pronunciation characteristics associated with this type of FAS. Several findings were considered essential in the diagnosis of psychogenic FAS: the psychological assessments as well as the clinical interview confirmed the presence of psychological problems, while neurological damage was excluded by means of repeated neuroimaging and neurological examinations. The type and nature of the speech symptoms and the accent fluctuations associated with the patient's psychological state cannot be explained by a neurological disorder. Moreover, the indifference of the patient towards her condition may also suggest a psychogenic etiology, as the opposite is usually observed in neurogenic FAS patients.

7.1. INTRODUCTION

Foreign accent syndrome (FAS) is a rare motor speech disorder that causes patients to speak their native language with an accent that is perceived as non-native by speakers of the same speech community. This 'non-nativeness' is the result of suprasegmental and/or segmental changes, which – according to the criteria proposed by Whitaker (1982) – are the consequence of damage to the central nervous system. Often, the etiology is stroke or brain trauma affecting the language dominant areas of the brain, e.g. the left (pre)frontal, temporal and/or parietal region, the rolandic and perisylvian area, as well as the insular region. Nevertheless, FAS has also been associated with other etiologies including MS (Villaverde-González et al., 2003; Chanson et al., 2009; Bakker et al., 2004), neoplasms (Masao et al., 2011; Tomasino et al., 2013; Abel et al., 2009) and vascular dementia (Paquier & Assal, 2007). Verhoeven & Mariën (2010a) argue that FAS is not only caused by (acute) neurological damage but it can also result from psychogenic issues. In psychogenic FAS, the accent is associated with a psychological/psychiatric disorder. Furthermore, Verhoeven & Mariën (2010a) also identified a mixed type in which FAS initially develops on the basis of a neurological disorder: this affects patients so profoundly that they further develop the accent in order to create the impression of a more authentic personality.

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The current study focuses on psychogenic FAS. For most of the psychogenic cases reported so far, a psychogenic cause was assumed because it was not possible to unambiguously identify a neurological disorder. Some authors have discarded the idea of psychogenic FAS because of diagnostic difficulties to objectify this condition (Gurd et al., 2001; Poulin et al., 2007). In some patients diagnosed with psychogenic FAS (repeated) brain imaging with CT or MRI revealed structural damage, but the speech problems were disproportionate in relation to the damage. Furthermore, in the majority of the psychogenic FAS cases symptoms were fluctuating, increasing in certain (social/emotional) contexts, diminishing or even completely resolving in others (e.g. Tsuruga et al., 2008, Haley et al., 2010, Jones et al., 2011, Van Borsel et al., 2005). Such a fluctuating course of symptoms is typical of speech and voice disorders of psychogenic origin (Avbersek & Sisodiya, 2010). When FAS is typified by these phenomena and associated with identifiable psychological problems (e.g. depression, familial history, suicidal ideation) a non-organic origin may be expected (Baumgartner, 1999; Roth et al., 1989; Tippett & Siebens, 1991, Baumgartner & Duffy, 1997).

7.2. BACKGROUND

In little over a century – counting from the first (anecdotal) FAS description by Pierre Marie in 1907 until July of 2014 – only 15 FAS cases with a presumed psychogenic origin have been reported (Critchley, 1964; Gurd et al., 2001; Reeves & Norton, 2001; Van Borsel et al., 2005; Verhoeven et al., 2005; Reeves et al., 2007; Poulin et al., 2007 = Roy et al., 2012, case 1; Tsuruga et al., 2008; Haley et al., 2010; Cottingham & Boone, 2010; Jones et al., 2011; Lewis et al., 2013; Polak et al., 2013). This study presents a new case of psychogenic FAS. Neuropsychological testing was carried out to assess a wide range of cognitive functions. The psychological state of the patient was evaluated by means of a series of psychodiagnostic tests, including symptom validity tests. Extensive neuropsychological investigations (Verhoeven et al., 2005; Poulin et al., 2007; Haley et al., 2010) and psychodiagnostic testing (Verhoeven et al., 2005; Cottingham & Boone, 2010) have only been occasionally reported in psychogenic case reports, although such an in-depth investigation is crucially important for accurate diagnosis and successful therapy (see also: Moreno-Torres et al., 2013). In addition, a perceptual analysis of the patient's most salient speech characteristics was carried out and an accent rating experiment was run to find out to what extent the patient's accent was considered non-native. Additionally, the listening panel was asked to indicate the mother tongue of the FAS speaker. Such experiments have previously only been reported in three studies (Di Dio et al., 2006; Verhoeven et al., 2013: rating and attribution experiment; Dankovičová & Hunt, 2011: rating experiment). We are convinced that perceptual assessment reinforces the diagnosis of FAS and it may provide new insights into the perceptual impression(s) created by FAS in the ear of the listener (Verhoeven et al., 2013).

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The patient gave written informed consent to report the medical data. All the tests reported below are part of the standard, clinical neurolinguistic work-up in patients with speech and language disorders at ZNA Middelheim general hospital. Speech recordings were also made to allow for better follow-up. The patient gave written consent to use recorded speech samples for the perceptual evaluation in a public environment.

7.2.1. Case presentation and medical history

SB is a 33-year-old, right-handed, monolingual French-speaking lady, originating from a village in the francophone Walloon part of Belgium near the Flemish border. She was raised in French and her parents were monolingual French-speaking Belgians. From a neurological perspective, growth and development were unremarkable. There was no family history of neurodevelopmental disorders or learning disabilities. She had always obtained normal school results and had an educational level of 12 years. She consulted the neurology department in November 2013 because of a 'Dutch or German-like accent', which she acutely developed approximately six months after she was hit by a car while crossing the street to deliver orders from the bakery where she worked as a saleswoman. A few months after the accident occurred, the patient mentioned an 'abrupt change of personality'. She considered her behavioral change as the cause for her sudden dismissal at work. There had been serious disagreements with colleagues, customers, as well as with her line manager. She was dismissed in June 2012. It was shortly after her dismissal that she developed a foreign accent.

The accident happened in December 2011. There had been no loss of consciousness. Apart from some superficial subcutaneous hematomas in the frontal and right peri-orbital region, clinical examination on admission to the hospital was normal. CT scan of the brain and spinal cord were normal. A diagnosis of minor head trauma was made. One week later, the patient started suffering from increasingly painful headaches (possibly a post-traumatic migraine, see: Weiss et al., 1991) and a desensitization of the scalp. She complained of vertigo and was hospitalized for three days. The clinical neurological examination on admission was normal. Laboratory investigations (blood and urine), EEG and CT were normal as well. She was diagnosed with a post-concussion syndrome, benign paroxysmal vertigo (positive Hallpike test) and a cervical trauma. Approximately one month later the symptoms were still present. She identified several regions of hyperaesthesia and anaesthesia in the facial area and the scalp. The vertigo had receded, but she complained of severe neck and shoulder pain. Approximately four months after the accident, she consulted a neurologist again. The clinical neurological examination and EEG revealed no abnormalities. During this visit, the patient mentioned that she felt she had become 'someone else' after the accident, with regular aggressive outbursts towards family, friends, strangers, and clients. The patient complained about attention deficits and permanent fatigue. She also mentioned that the intensity of the accent was fluctuating: the accent was heavier when she was tired.

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Due to the persistence of her complaints with respect to her accented speech and memory, the patient was referred to hospital for additional radiological examinations. In November 2012, she underwent a saggital T1-weighted and axial FLAIR, diffusion, SWI, proton density and T2-weighted MRI of the head, a coronal FLAIR MRI perpendicular to the axes of the left and right hippocampi, as well as an angio-MRI of the brain and 3D TOF of the circle of Willis. The qualified radiologist reported that all acquisitions were normal.

In November 2013, she consulted our department because of her persistence of the accent change and cognitive complaints (attention problems and episodes of confusion). At a linguistic level she suffered from word-finding difficulties and morphological problems related to article-noun agreement (she did not differentiate between the masculine and feminine forms of the definite article). According to her, listeners had the impression that she spoke with a Dutch accent. Her previous customers, for instance, had perceived her as a native Dutch-speaking Belgian and repeatedly asked her why she spoke French instead of 'Flemish' (the Belgian variant of Dutch) (see: Verhoeven, 2005). She still suffered from behavioral changes and avoided social contact with her family and friends because of a lack of interest on her part. Yet, she was looking for more excitement in life, as well as a more frivolous, out-going lifestyle. She said she was deeply bored. In addition, a number of depressive symptoms were mentioned including apathy, loss of drive and initiative, and mood-swings.

7.2.2. Neuropsychological testing

The first neuropsychological assessments were carried out approximately one year after the accident in January 2012 (see Appendix D1 for an overview of the results). The test battery consisted of the Wechsler Adult Intelligence Scale-IV (WAIS-IV; Wechsler 2011, French Ed.), the d2-test (Brickenkamp & Zillmer, 1998), the 'Barrage de Zazzo' (Zazzo, 1974), the Stroop Test (Stroop, 1935), the Wisconsin Card Sorting Test (WCST; Grant & Berg, 1948), and the California Verbal Learning Task (CVLT, Delis et al., 2000). Repeated neuropsychological testing in 2014 consisted of the Wechsler Memory Scale – Revised (Wechsler, 1987), the Boston Naming Test (Kaplan et al., 1983); the Trail Making Test (Reitan, 1958) and the d2-test.

A full scale IQ (FSIQ) of 105 was found with a significant discrepancy of 24 IQ-points between the verbal (96) and performance IQ level (120). All subtest scores were within the normal range. Executive functions (mental flexibility, frontal problem solving) were tested by means of the Stroop and the WCST. She obtained a normal result on the WCST, but depressed scores on the Stroop (slowed processing in the color naming condition (Z-score= -1.5 SD), interference condition (Z-score= -1,6 SD) and flexibility condition (Z-score= -1.7 SD). Tests measuring sustained visuo-motor and selective attention (d2-test in 2012/2014 and the 'test de barrage de Zazzo') were performed at a slow pace. Scores for total items treated for the d2-test (2012: Z= -3.08 SD; 2014: Z= -2.44 SD) as well as the total items corrected (2012: Z= -2.94 SD; 2014: Z= -2.20 SD) were in the

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pathological range. As shown by the CVLT, verbal memory was intact, the patient obtained borderline results for the 'total recollection' (5 trials) of List A (Z-score= -1.49 SD). On other subtasks of the CVLT she obtained normal results (+1 SD: Cued recall A, Delayed recall A, Cued delayed recall A, Recognition).

In 2014, a significant discrepancy between a very superior visual memory index (= 133) and clinically deficient verbal memory index (= 74; -1.7 SD) was found on the WMS-R. As reflected by a general attention index of 70 (-2 SD), the WMS-R tasks scores were in the deficient range. The Trail Making Test (part A and B) disclosed low average visual search (< pct. 10) and mental flexibility (pct. 20). Sustained visuo-motor attention scores were within the defective range. Performance on the BNT was normal. Overall, the data for the test session in 2014 were in line with the results obtained in 2012.

7.2.3. Psychodiagnostic assessment

The psychodiagnostic assessment consisted of an interview with an experienced clinical psychologist (LDP), which was followed some time later by a session during which the patient was asked to respond to a series of standardized questionnaires. These questionnaires were completed at the hospital, without the help of the examiner. Testing included the Minnesota Multiphasic Personality Inventory-2 (MMPI-2: Butcher et al., 1989); the Defense Style Questionnaire (DSQ-60: Thygesen et al., 2008); the Rotter Incomplete Sentences Blank (RISB-FR: Rotter et al., 1992); Beck Depression Inventory-II (BDI-2: Beck et al., 1996), Pathological Narcissism Inventory (PNI: Pincus et al., 2009; French version: Diguer et al., 2014) and the Narcissistic Personality Inventory-40 (NPI-40: Raskin & Hall, 1979).

Furthermore, symptom validity and self-presentation tests were carried out by means of the List of Indiscriminate Psychopathology (LIPP: Merten & Stevens, 2012), and the Supernormality Scale (SS: Cima et al., 2003). The LIPP is an experimental questionnaire, which measures calibration problems. It consists of questions addressing pseudo and real symptoms (Merckelbach et al., 2013). Malingering participants are in doubt as to which symptoms they can report and which ones they cannot. The SS is a questionnaire, which evaluates deception or denial under the guise of giving socially desirable answers (Cima et al., 2003).

During intake the patient gave evidence of disinhibition which mainly manifested itself as laughing without reason, Witzelsücht and inappropriate comments. The patient was reticent and maintained a (psychologically immature) defensive attitude throughout the entire interview. Her thoughts were preoccupied by frustration about her own situation. The interview was dominated by her feelings concerning her increased impulsiveness, aggressiveness and apathetic demeanor vis-à-vis her family, former boss and colleagues. The examiner noticed that a topic that rendered her frustrated led to an emotional breakthrough during which she lost the 'Dutch/Flemish-like' accent. The patient's interview contained numerous contradictions (e.g., stating at first that she was a very lively, out-going person, but when asked later what she did during

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the day, she answered that she sat in a chair as all personal contact bored her and conversations with others – even friends – were too difficult and tiring). The description of her emotional and family life remained superficial and prosaic. The interview revealed increasing relational problems. The relationship with her husband left her 'unaffected' and relationships with friends, family and relatives were unstable, marked by serious altercations in which she responded unpredictably.

She confirmed egocentric and narcissistic tendencies. It was not possible to detect signs of perceptual aberration or other florid psychotic symptoms. A few weeks after the interview, a series of standardized psychodiagnostic tests were administered. Symptom validity and self-presentation tests, such as the List of Indiscriminate Psychopathology and the Supernormality Scale, did not yield indications for (conscious or unconscious) manipulation. Personality testing indicated a wide, undifferentiated personality disturbance. Interestingly, scores on both narcissism measures (NPI and PNI) were at most extreme upper ends, which is consistent with her answers during the clinical interview. A thymic disturbance and affective lability were objectified (APA, 2000; DSM-IV-TR, Axis I), but test results did not equivocally point towards a well-defined personality disturbance. Clinically, however, the patient gave clear indications of highly dependent, histrionic and borderline personality characteristics (APA, 2000; DSM-IV-TR, Axis II). On a psychodynamic structural level, she was considered to have a borderline personality organization level of functioning (Kernberg, 1984), because of an immature defensive functioning, intact reality testing, but severe lack of personality integration. This is relevant in relation to (interpersonal) acting out and poor bodily representation. The overall clinical presentation seemed chronic, pervasive and well established throughout her psychic development.

7.2.4. Perceptual analysis of spontaneous speech sample

A post-morbid speech sample was recorded in November 2013. It consisted of 5 minutes of video-recorded spontaneous speech, which was selected from an interview with the patient. In this interview she talks about her accent change and her relational and professional problems. This sample consisted of 644 words (including filled pauses). The patient also provided two (short) pre-morbid speech samples consisting of 43 and 26 seconds of conversational speech dating from April and July 2011, i.e. approximately half a year before the accident. When comparing pre- and post-morbid speech samples a number of striking differences were found. The first one was a very strong trilling aspect when realizing the uvular [R]. The trill is too excessive for French, and is more typical of the one in German and some regional variants of Dutch (36/644). According to Van de Velde & Van Hout (1999, p. 178) "realizations of /r/ in standard Dutch until recently were the trilled realizations [R] and [r], with the uvular trill gaining in frequency and prestige especially in the Netherlands (Van Haeringen, 1924; Zwaardemaker & Eijkman, 1928; Blancquaert, 1934; Hol, 1951; Damsteegt, 1969; Mees & Collins, 1982; Vieregge & Broeders, 1993), but recently also in Flanders (Rogier, 1994)". For German

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this variant has been described as the most common allophone (Hall, 1993): the uvular trill-R constitutes a free (dialectal) variant of /r/, existing alongside the approximant /ɹ/ (see Hall, 1993; Schiller, 1998). The excess trilling is particularly common in a prevocalic position (*r*aconter, *r*enverse, t*r*aite, …: 27x), less frequent in intervocalic position (di*r*ect: 1x) and postvocalic position (renver*s*e, qua*r*t, …: 8x).

On the suprasegmental level, speech rate and articulation rate were particularly slow (speech rate: 2.67 syll/sec, articulation rate: 3.813 syll/sec). Avanzi et al. (2012) found a mean speech rate of 4.7 syll/sec (SD: 0.7) and an articulation rate of 5.6 syll/sec (SD: 0.6) for Belgian French of the Tournai region; the region our patient originated from. Melody and intonation appeared normal. In order to analyze rhythm, the Pairwise Variability Index was calculated (Low et al., 2001). Vocalic PVI amounted to 54.3. This is considerably higher than the accepted value for French (43.5), and is more in the range of the stress-timed languages, such as English (57.2), or German (59.7). However, the value is substantially lower than 65.5, which is the reference value for Dutch (Grabe & Low, 2002). It is also worth mentioning that the patient did not articulate any liaisons, a phenomenon by which a latent word-final consonant preceding a word starting with a vowel becomes audible. Our patient failed to realize this connection for 'c'est arrivé' and 'tout est important'. Moreover, she did not articulate the elision[26] in 'j'entends' (pronounced as '*je* entends*')

Grammar was perceived to be more simplistic than would be expected from a native-speaker of French. Sentences were perceived to be very short. At the morphosyntactic level the patient omitted the article 'le' (1/644) as well as 'de' in 'là *de*dans', which was realized as 'là dans' (2/644). In addition, the patient made six morphological errors against the definite article. In 5 instances, the patient used the masculine definite article instead of the female form (la même chose → *le* même chose; la tête → *le* tête; à l'hôpital → au hôpital; ma maison→ *mon* maison; la pire chose→ *le* pire chose; la chose → *le* chose).

7.3. PERCEPTUAL ASSESSMENT OF THE FOREIGN ACCENT

7.3.1. Aims

The foreign accent of the patient was assessed by a listening panel which listened to speech stimuli of the patient that were mixed with those of a native speaker of French and three non-native speakers with a clear foreign accent. The listening panel was re-

[26] In English, the term elision is sometimes used as a synonym for deletion (e.g. Miller et al., 2006). For current article, we make a distinction between a 'deletion' and an 'elision' (French: *'élision'*), which is the "the suppression of a word-final vowel preceding a word starting with a vowel" (in spoken French this can refer to actual vowels, or the latent word-initial '*h*' preceding a vowel – with a few exceptions) (Schane and Filloux, 1967, p. 37, *our translation*).

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quired to rate the degree of foreignness and they were asked to identify the mother tongue of each of the speakers. The ratings provide additional support for the diagnosis of FAS, whereas the accent attribution gives an indication of whether naive listeners are able to perceptually identify the mother tongue of native (including the FAS patient) and non-native speakers of French. Furthermore, there was an interest to investigate whether there would be any differences between the FAS patient, the true non-native speakers and the native speaker of French.

7.3.2. Methods

7.3.2.1. Materials and samples

Thirty students of French linguistics were recruited at the Université Libre de Bruxelles (ULB) in Brussels (age: 16-24, mean age: 20 years, 12 male and 18 female) and they were asked to rate the degree of 'foreign-ness' of five speakers and to determine their native language. The students had no formal experience with speech and language pathology.

The stimuli for this experiment were taken from the intake interview, in which the patient explains what had happened to her (accident), and elaborates on her relational and professional problems. From this interview, 6 words, 3 phrases and 6 sentences were chosen (see also: Dankovičová & Hunt, 2011). Care was taken that (a) the medical status of the FAS patient could not be derived from the stimuli and (b) the stimuli did not contain any morphological mistakes (as this could possibly influence the ratings of the listener panel). Stimulus selection was carried out by means of PRAAT, version 5.4 (PRAAT for Mac; Boersma & Weenink, 2014).

7.3.2.2. Speakers

The speakers in this experiment were the FAS patient and four control speakers (**Table 7.1**) who were matched for gender with the FAS patient. The mean age of the controls was 35 years and 10 months, with an age range from 27 to 48 years old. Two speakers were Belgian but one was French-speaking and the other was Dutch-speaking (or 'Flemish'; see also: Verhoeven, 2005). A third control subject spoke both Dutch and (American) English, as she was born in the USA, but moved to the Netherlands one year later. She was raised in English, but her education as of the age of 3 had been entirely in Dutch (100% immersion; early bilingual; see also: Bhatia & Ritchie, 2013). She no longer had contact with relatives in the USA and lived alone in the Netherlands. She considered Dutch to be her dominant language. The fourth speaker was a Russian female. No attempt was made to match the accents to those that had been informally reported for the FAS patient. It was regarded likely that most listeners were familiar with the foreign accents of the control speakers. The control speakers read the 15 stimuli that had been selected from the speech of the FAS patient. The stimuli were recorded by means of a

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Marantz Professional PMD 661 portable recorder and manipulated for the purpose of this experiment via PRAAT (version 5.4, 2014).

Table 7.1. Overview of the demographic characteristics of the FAS patient and the healthy, matched controls, including an indication of the level of French (CEFR= Common European Framework of Reference for Languages). *Control 3 moved to the Netherlands one year after she was born. She was raised in English and learned Dutch as of the age of 3. Her education (immersion, 100%; early bilingual) has been entirely in Dutch.

Nature	Gender	Age	Country of Birth	Mother tongue	Level in French (CEFR)
FAS	F	33	Belgium	French	_
Control 1	F	36	Belgium	French	_
Control 2	F	48	Belgium	Dutch (Flemish)	B2
Control 3	F	27	United States of America*	English / Dutch (Netherlands)	B2
Control 4	F	35	Russia	Russian	A2+ / B1

7.3.2.3. Stimuli and assessment

The perception experiment contained a total of 75 stimuli, i.e. 15 stimuli x 5 speakers. Each presentation block consisted of one stimulus read by the five different speakers. The order of the speakers differed for each block (in pseudo-random order). The stimuli were separated by a 15 sec. pause to provide time for listeners to record their judgments. Total duration was 26 min. 26 sec. The stimuli were played to the listeners in open field at their institution. The instructions to the test were given orally to the listening panel, but they were also able to read them. Raters provided demographic information (age, gender, country of origin, time in Belgium - if not born here, mother tongue, and other spoken languages including an indication of proficiency) in a short questionnaire. For the experiment, they were asked to first rate the 'foreign-ness' of the speaker on a scale from 1 to 7. This scale is to be interpreted as a continuum ranging from 'definitely *not* a native speaker of French' (=1) to 'definitely a native speaker of French' (=7). If their response was anything other than 7, they were asked to indicate the mother tongue of the speaker (second part).

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7.4. RESULTS

7.4.1. Statistical analysis of the accent rating experiment

The data were processed statistically in SPSS version 22 (IBM corp., 2013). First, inter-rater reliability was tested for each speaker by calculating the intraclass correlation coefficient (ICC). A two-way random model was chosen, as each item was assessed by each of the 30 raters and raters represented a randomly selected sample. Data were checked for agreement implying that systematic differences between raters were taken into account. For FAS: ICC(2,30)=0.94, French: ICC(2,30)=0.903, Dutch(Be): ICC(2,30)= 0.955, English/Dutch(Nl): ICC(2,30)= 0.959, and for Russian: ICC(2,30)= 0.523.

Table 7.2 provides a summary of the descriptive statistics including means, standard deviations, minima and maxima, range as well as interquartile range for each of the five speakers. Based on the means ($\bar{x}$) as well as median (M) it is clear that the FAS speaker is situated roughly in the middle of the seven-point scale ($\bar{x}$ = 3.791; σ =2.318 and M=4). The standard deviation was high, which indicates that the raters may have experienced some difficulty identifying the accent.

Table 7.2. Overview of mean, median, standard deviations, minimum, maximum, range and interquartile range for the scores attributed to each speaker on a seven-point scale: 1= Definitely not a native speaker of French; 7= Definitely a native speaker of French. Min= minimum, Max= maximum, SD= Standard Deviation.

SPEAKER	Mean	Median	SD	Min	Max	Range	Interquartile range
FAS	3.791	4.000	2.318	1.000	7.000	6.000	5.000
FRENCH2	6.098	7.000	1.675	1.000	7.000	6.000	1.000
DUTCH (Be)	3.138	3.000	2.161	1.000	7.000	6.000	4.000
ENGLISH/DUTCH (Nl)	3.011	2.000	2.219	1.000	7.000	6.000	4.000
RUSSIAN	1.407	1.000	0.913	1.000	7.000	6.000	0.000

Application of the Kolmogorov-Smirnov test indicated that the data were not normally distributed (Kolmogorov-Smirnov: $p < 0.01$). Hence, non-parametric testing was applied. A Kruskall-Wallis *H* test was carried out to test whether there was a significant difference between the scores attributed to the different speakers. Results for the Kruskall-Wallis *H* test indicated that this was the case: $H(5)= 1393.60$, $p < 0.0001$. However, additional Mann-Whitney *U* tests (see **Table 7.3**) were carried out to identify the speakers who differed significantly from each other and who did not. All speaker

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differences were significant ($p < 0.0001$), except for one: Dutch (Be) and English/ Dutch(Nl) ($p > 0.003$: Bonferroni correction; $p = 0.290$).

Table 7.3. Overview of the inter-speaker comparisons (Mann-Whitney U-tests)

SPEAKER COMPARISON	N stimuli	MEAN RANKS	SUM OF MEAN RANKS	MANN WHITNEY U	WILCOXON W	Z	*p*
FAS	450	322.88	145294.50	43819.500	145294.500	-15.497	0.000
FRENCH	450	578.12	260155.50				
FAS	450	487.20	219240.00	84735.00	186210.00	-4.326	0.000
DUTCH(Be)	450	413.80	186210.00				
FAS	450	494.07	222331.50	81643.500	183118.500	-5.154	0.000
ENGLISH/ DUTCH(Nl)	450	406.93	183118.50				
FAS	450	587.61	264424.50	39550.500	141025.500	-17.102	0.000
RUSSIAN	450	313.39	141025.50				
FRENCH	450	606.39	272845.00	31100.00	132575.000	-18.728	0.000
DUTCH(Be)	450	294.61	132575.00				
FRENCH	450	604.38	271972.00	32003.000	133478.000	-18.576	0.000
ENGLISH/ DUTCH(Nl)	450	296.62	133478.00				
FRENCH	450	659.11	296598.00	7377.000	108852.000	-25.523	0.000
RUSSIAN	450	241.89	108852.00				
DUTCH(Be)	450	459.37	206717.50	97257.500	198732.500	-1.059	0.290
ENGLISH/ DUTCH(Nl)	450	441.63	198732.50				
DUTCH(Be)	450	558.65	251391.50	52583.5	154058.500	-13.864	0.000
RUSSIAN	450	342.35	154058.50				
ENGLISH/ DUTCH(Nl)	450	547.76	246494.00	57481.000	158956.00	-12.628	0.000
RUSSIAN	450	353.24	158956.00				

A correspondence analysis was performed to get a two dimensional image of the strength (distance) of the associations between rating and speakers, based on frequency counts (**Table 7.4**: correspondence table) (**Fig. 7.1**). This showed that the associations between the native French speaker and rating '7' were particularly strong. The FAS speaker was situated more towards the higher ratings (4, 5, 6, 7) than, for instance, both native Dutch speakers and even markedly more so than the Russian speaker (strongly associated with rating '1'), who clearly occupied a more isolated position on the two-dimensional plot.

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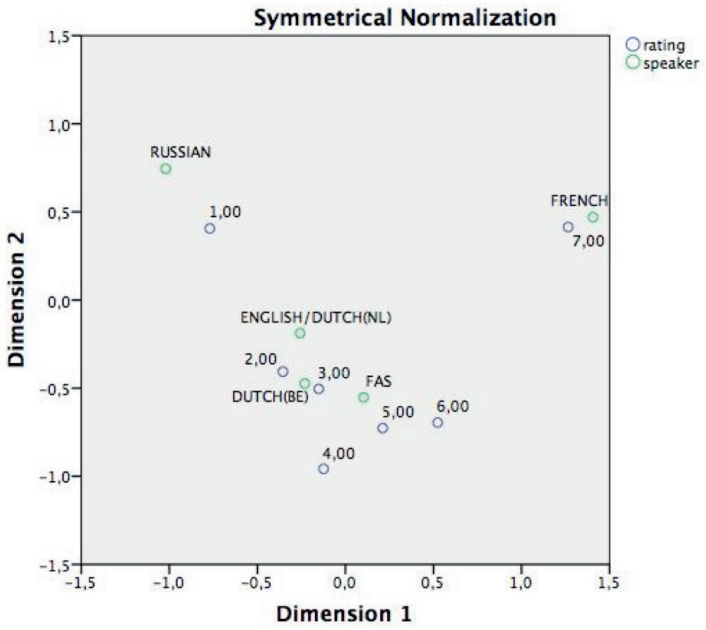
Table 7.4. Correspondence table with frequency data for the different speakers. 1= Definitely not a native speaker of French; 7= Definitely a native speaker of French

Correspondence Table

Speaker	rating							
	1	2	3	4	5	6	7	Active Margin
FAS	118	62	40	42	45	50	93	450
FRENCH	18	15	21	10	34	41	311	450
DUTCH(BE)	165	59	51	38	51	33	53	450
ENGLISH/DUTCH(NL)	183	62	47	35	36	22	65	450
RUSSIAN	349	53	27	11	8	1	1	450
Active Margin	833	251	186	136	174	147	523	2250

Figure 7.1. Correspondence analysis, displaying the associations between speakers and rating in a two-dimensional plain. As can be derived from the figure, both the Russian and the French speaker maintain an isolated position in the plain and are associated with opposite extremes of the continuum. The English/Dutch(Nl), Dutch(Be) and FAS speaker on the hand, are all grouped around the center ratings: 2,3,4, and 5.



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7.4.2. Mother tongue identification

It appeared that only 50% of the raters (*n*=15/30) had indicated the mother tongue of each speaker for each stimulus. Nine raters were female, and six were male (age range: 16-23 years; mean age: 19 years). **Figure 7.2** shows the different accents associated with the different speakers. Exact numbers can be found in **Table 7.5**.

Table 7.5. Overview of the mother tongues (rows) associated with each of the speakers (columns). Languages pertaining to the same language family have been grouped together

	FAS	FRENCH	DUTCH	ENGLISH/ DUTCH	RUSSIAN
French	61	214	39	35	4
Spanish	21	0	8	33	19
Italian	21	1	7	10	32
Portuguese	5	0	0	2	22
Romanian	4	0	0	0	3
Dutch	46	5	120	63	25
English	8	0	10	19	16
Danish	0	0	1	0	0
Norwegian	0	0	1	0	0
German	10	0	20	23	5
Luxembourgish	0	0	1	0	0
Greek	1	0	0	1	2
Polish	8	0	0	10	27
Russian	3	2	2	7	64
Slavic	1	0	0	0	1
Chinese	1	0	0	0	0
Japanese	3	0	1	2	0
Vietnamese	0	0	0	1	0
Asian-sounding accent	1	0	0	1	0
Hungarian	0	0	1	1	1
Estonian	1	0	0	0	0
Lithuanian	0	0	0	1	0
Turkish	0	0	0	0	0
Basque	0	1	0	0	0
Unidentifiable	30	2	14	16	4
TOTAL	**225**	**225**	**225**	**225**	**225**

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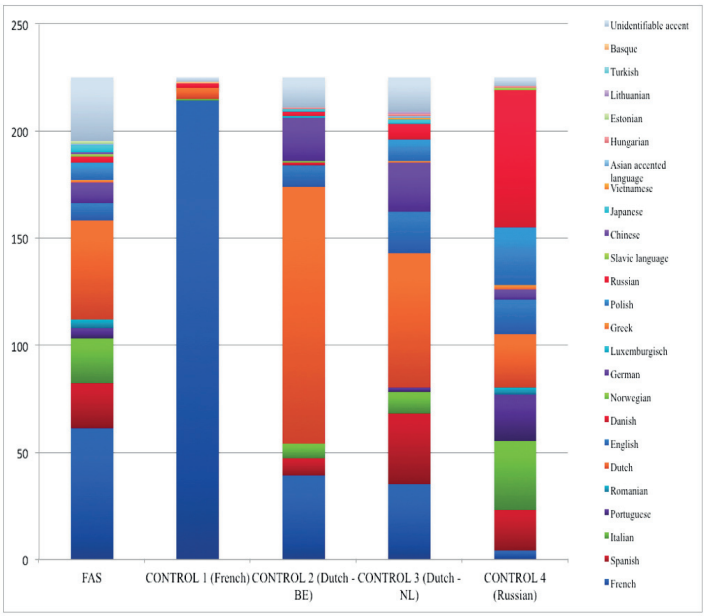
In general, the FAS patient was less often identified as 'French' (*n*=61/225; 27.1%) than as a speaker of other languages (72.9%). However, the other languages attributed to the FAS patient were most often Romance languages (Spanish: *n*=21; Italian: *n*=21; Portuguese: *n*=5; Romanian: *n*=4; *n*=112/225; 49.8%). Still, she was identified as Dutch in 21.3% of the stimuli (*n*=48/225), and as German in 4% of stimuli (*n*=10/225). The FAS patient was less often identified as 'French' than the native French speaker (*n*=214/225; 95.1%), which corroborates the findings for the first part of the study. Hence, there seemed to be a clear difference in the perception of the FAS patient and the non-impaired French control speaker. The Dutch (Be) speaker was associated with 'Dutch' in 53.3% (*n*=120/225) of the stimuli, whereas for the English/Dutch(Nl) speaker this was 28% of the stimuli (*n*=63/225). In 8% of the stimuli she was associated with 'English' (*n*=19/225). The Russian speaker was correctly associated with her native language in 28.4% (*n*=64/225) of the stimuli.

Figure 7.2. Graphical overview of the stratification of the different mother tongues associated with the different speakers



Interestingly, the accent stratification was most diverse for the FAS patient (16 different mother tongues were associated with her stimuli). For the other speakers, the number of attributed accents was: English/Dutch(Nl): 15; Russian: 13; Dutch (Be): 12; and French: 5. Equally interesting to note is that the accent of the FAS patient could not be identified in 30 instances: this is considerably more often than for the other control speakers: French: 2; Dutch (Be): 14; English/Dutch (Nl): 16; Russian: 1.

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7.5. DISCUSSION

This article discusses the case of a patient who developed FAS in the absence of demonstrable damage to the central nervous system. No structural damage was visible on repeat CT and MRI of the brain. Repeat neurological and neurophysiological examinations were normal. An in-depth psychodiagnostic work-up was carried out (a) to confirm the existence of psychological issues and (b) to identify a possible psychiatric disorder. Unfortunately, testing did not reveal a clearly delineated disorder on either axis I or II of the DSM-IV-TR (APA, 2000). Test results were, however, indicative of a highly dependent, hysterical and borderline personality. Although psychological problems were considered persistent and chronic, there were several elements in the clinical interviews that could corroborate the hypothesis of a psychogenic origin of the accent change.

First, the accent diminished whenever there was a psychological breakthrough during the clinical interview (Avbersek & Sisodiya, 2010; see also: Keulen et al., 2016b[27]). More specifically these episodes occurred when the patient talked about her relational problems, issues at her former workplace and the fact that she no longer had a job at the moment the interview took place. Interestingly, the *negative* impact of emotional disequilibrium, feelings of stress and/or anxiety on the recovery process has previously been established for neurological speech and language disorders (see also: Cahana-Amitay & Martin, 2015). In contrast, the current patient seemed to *benefit* from these emotional triggers.

Second, there was a correspondence between the culmination of disputes with her line manager, which ultimately led to her dismissal, and the onset of the accent: both occurred approximately six months after the accident.

Third – and related to prior argument – the increased emotional lability and hysteric symptoms may have been reinforced by the adverse life events that had marked her life in rapid succession: the car accident, the accent shift, the dismissal, and the relational problems. According to Avison & Turner (1988) the relationship between adverse life events and psychological distress is often underestimated. According to Charles et al. (2013) even naturally occurring daily stressors or minor affective experiences can have a far-reaching impact on mental health (p. 739). It is important to note that at the time we saw the patient, she had been unemployed for about a year and a half and had marital problems.

The patient had repeatedly complained about (sustained) attention, memory and slow cognitive processing. These complaints were confirmed by neuropsychological test results: the patient showed evidence of impaired processing on the cognitive tasks appealing to working memory, attention, and executive function. These complaints have been noted regularly in psychogenic FAS patients (Poulin et al., 2007 = case Roy et al., 2012; Cottingham & Boone, 2010; Jones et al., 2011) and have more generally been

[27] In this article, another case of psychogenic FAS is presented. The patient suddenly lost her accent during a temper tantrum.

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associated with somatic disorders (Demir et al., 2013; Trivedi, 2006; Niemi et al., 2002). However, studies claiming such an association have been the subject of scientific scrutiny, because hardly anyone administered symptom validity tests to their participants prior to inclusion. Delis & Wetter (2007) suggest that patients with psychogenic disorders may exaggerate cognitive deficits, due to external (medico-legal reasons, treatment), internal/interpersonal incentives (in order to sustain a dependent relationship with specialist or other) or even for unspecified reasons ('not otherwise specified'). The current patient completed symptom validity tests, which turned out negative for malingering and feigning. Moreover, neurocognitive testing was carried out on two occasions (two years apart, both post-morbid). This is crucial, as significant underperformance or inconsistencies in cognitive test scores or profiles across repeated evaluations would be considered indicative of a feigned cognitive deficit.

In the case of our patient the profile seems mostly consistent with a post-concussion cognitive syndrome after a minor head trauma. The objectively attested cognitive deficits and the negligence of the cognitive complaints after prior examinations might also have contributed to the development of the FAS.

On a linguistic level, the patient's speech was characterized by the realization of the uvular R with a marked, atypical trill and occasionally, she deleted phonemes. Furthermore, the patient spoke at a very slow speech rate and had a speech rhythm that was qualified as stress-timed, whilst French is a syllable-timed language (Grabe & Low, 2002). The segmental and suprasegmental alterations noted for this patient do not seem to be restricted to a psychogenic population: all have been attested for neurogenic FAS patients as well. However, the isolated, morphological deficits, which irregularly affected articles, and the occasional pronunciation deficits affecting liaisons and elisions (phenomena typifying French) seemed incredible. The grammatical deficit is very different and less substantial than the agrammatism and paragrammatism encountered in aphasics, for instance. Some degree of conscious or subconscious manipulation cannot be ruled out. Incredible grammatical disorders of the like have previously been reported in other psychogenic cases (e.g. Van Borsel et al., 2005; Cottingham & Boone, 2010).

Some speech characteristics might have been consistent with the impression of a Dutch or German accent. However, results of the listening experiment suggest that the patient was perceptually situated midway between a true non-native speaker of French and a native speaker of French. This finding is in line with what has been found in the experiments of Verhoeven et al. (2013) and Di Dio et al. (2006). However, their methodology differed from the current one in the sense that they did not select words and sentences in pseudo-random order, but provided raters with spontaneous speech samples. The methodology in the present experiment was more comparable to the approach of Dankovičová & Hunt (2011), who used single words and phrases. As far as the identification of the linguistic background of the speakers is concerned, it was found that the FAS patient was associated with French in only 27.1% of the stimuli. The French-speaking control subject on the other hand was almost always recognized as French

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(95.1%). The patient also demonstrated the most diverse association patterns regarding her native language. The 'uncertainty' expressed in the first part of the experiment (M= 4) compares well with the second part of the study: the patient was associated with 16 different possible native languages, and for 13% of the items, the mother tongue could not be identified. Furthermore, the hypothesis that the patient was perceived as Dutch or German was not entirely confirmed, as most listeners still perceived her as being a native speaker of a Romance language (for 49.8% of the stimuli, including French, Italian, Spanish, Portuguese, and Romanian; in comparison: Germanic languages, including English, Dutch and German: 28.4% of the stimuli).

Remarkably, the patient did not seem bothered by the accent change at all. Nevertheless, there were clear problems at the cognitive-behavioral and psychological level (mentioned above). Moreover, she was not keen to be treated for the condition. Rather, she wanted to show off with it. She did not seem to be overtly concerned about her symptoms. This is unlike what is mostly seen in neurogenic patients, who are emotionally and psychologically affected by FAS (Miller et al., 2011). In fact, to the best of our knowledge, there are only two other reports (Laures-Gore et al., 2006; Tailby et al., 2013) in which it was mentioned that patients were almost completely indifferent to the negative implications. These cases were classified as 'mixed FAS' by Verhoeven & Mariën (2010a): these patients further optimize their accent and often start to use words of the language, which is suggested by their accent in order to create a more authentic personality. The use of foreign-sounding words or a more formal language variant has also been noted for psychogenic FAS patients (Poulin et al., 2007, Reeves & Norton 2001, Polak et al., 2013; see also: Reeves et al., 2007, case 3). The processes that invoke this kind of change in language use, still remain to be clarified. For neurogenic cases, some positive associations have also been noted. According to some patients, living with FAS opened new horizons. However, in the longer term, the negative perceptions from others, the hybrid identity, a loss of sense of belonging, a breakdown of relationships, and the incapacity of medical staff to explain the change all lead to frustrations (Miller et al., 2011).

Patient coping strategies, psycho-emotional and social implications have generally been underreported in the literature about both psychogenic and neurogenic FAS (for neurogenic patients: Miller et al., 2006, Miller et al., 2011; Munson, 2005; Moreno-Torres et al., 2013). Future research should identify and study the effects of this syndrome at the personal and inter-personal level to allow for a full rehabilitation of both speech profile and psychological well-being.

7.6. CONCLUDING REMARKS

Only a handful of putative psychogenic FAS cases have been described in the literature and many researchers have been hesitant to conclude to an underlying psychogenic etiology. Although it is hard to provide evidence for a direct causal link between the

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psychological factor in play and FAS, ample evidence exists that the FAS symptoms (and their course) in this patient are of a psychogenic nature: (1) clear absence of (visible) neurological damage or clinical evidence for a neurological disorder, in conjunction with (2) the presence of psychological and psychiatric factors, (3) the timing of the onset of the accent change, (4) the fluctuating symptom course, (5) irregular and incredible morphological mistakes occurring in a short sample of spontaneous speech, and the fact that (6) the patient was unconcerned by the change of accent. As most of the psychogenic FAS cases were published in the last decade, reports of cognitive-behavioral deficits such as the ones displayed by the current patient are becoming increasingly important with a view to the development of the proper therapeutic approaches for this psychogenic FAS population.

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CHAPTER 8

Neologistic Jargon in a case of Psychogenic Foreign Accent Syndrome

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ABSTRACT

A 28-year-old, monolingual, native Dutch-speaking woman developed Foreign Accent Syndrome (FAS) a few weeks after a fall down the staircase. In addition to FAS, her oral output was interspersed with German(-like) words and structures (language mixing and switching). Increased stress, fatigue or emotional load caused an aggravation of the speech symptoms and induced a neologistic jargon speech. This phenomenon was never before observed to co-occur with FAS. Apart from a borderline low intelligence, formal psychodiagnostic tests did not identify a clear disorder. The clinical neurolinguistic tableau was strongly indicative of an underlying psychogenic etiology.

8.1. INTRODUCTION

Healthy polyglot individuals automatically employ the correct language depending on the pragmatic/communicative context. Mixing (e.g. within utterance mixing of different languages) and switching of languages or codes (i.e. between languages or codes in alternating utterances) can occur when the speaker is in an on-going acquisition process, which is the case for young bilingual or polyglot children (Redlinger, 1980; Meisel, 2000). However, in the process of acquisition, adolescent or adult second language learners can equally demonstrate switching and mixing behavior (unbalanced bilinguals; Duran, 1994). In all these cases, the language mixing and switching has been argued to be the consequence of failed neural suppression of the language that was not targeted (e.g. for vocabulary the 'lexical competition'-phenomenon occurs, see also Abutalebi & Green, 2007). The efficiency of the inhibition mechanism depends on several factors such as the degree of proficiency, age of acquisition and computational load of the languages. These are hypothesized to be of influence on the strength of the induced plastic changes in the neural networks in the brain subserving for instance cognitive control, which is of utmost importance in this process of language inhibition (Perani & Abutalebi, 2005; Hernandez, 2009). Failed inhibition in these learners will not be interpreted as 'pathological', as errors are explicable within the dynamic framework of the aforementioned parameters.

Pathological language mixing and switching refer to the process of blended language use in bilingual or polyglot aphasic patients (Aglioti, 1993,1996; Abutalebi et al. 2008; Mariën et al. 2005; Ansaldo et al., 2010). Some researchers have argued that the basal ganglia play a crucial role in both language switching and mixing (Mariën et al., 2005; Aglioti et al., 1993), whereas others have argued that switching in particular is mainly subserved by frontal circuits (Fabbro et al., 2000; Hernandez et al., 2001) and mixing by the temporoparietal lobe (Fabbro et al., 2000).

Although typically described in the context of aphasia (e.g. Aglioti et al., 1996; Mariën et al. 2005), pathological language mixing and switching has been observed in

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a handful of cases with Foreign Accent Syndrome (FAS; Verhoeven et al., 2005; Reeves & Norton, 2001; Laures-Gore et al., 2006; Reeves & Norton, 2007). In all these cases pathological language mixing and switching occurred in the context of an underlying psychogenic disorder.

Jargon speech is usually associated with acquired aphasia (e.g. Hillis et al., 1999; Rohrer et al. 2009) and can be of a variable nature: semantic (language devoid of content); phonemic (production of words phonemically related to target), or neologistic (words not phonemically related to the target word) (see Rohrer et al., 2009). The patients are usually unaware of their mistakes. In contrast to '*schizophasia*', or the "meaningless mixture of words and phrases characteristic of advanced schizophrenia" (Paradis, 2008, p. 201) that can wax and wane, the patients suffering from jargon aphasia retain manifest expressive problems (Rohrer et al., 2009). The language disorder does not fluctuate, and the neurological symptoms will demonstrate a more linear resolution in case of remission.

Here we describe a 28-year-old native Dutch-speaking woman who developed psychogenic FAS a few weeks after a fall down the staircase. The patient had fluctuating FAS symptoms, and suffered from pathological language mixing (German), with neologisms. Her speech sometimes evolved to an incomprehensible neologistic jargon, a phenomenon never before reported in the literature on FAS.

8.2. CASE STUDY

The presented work-up integrally fits the standard clinical, neurocognitive work-up of patients with speech- and/or language disorders at ZNA Middelheim hospital. The patient provided written informed consent to report data from her medical file according to hospital regulations and standards. She gave a written permission to use speech samples for the linguistic analyses and to publish this article.

8.2.1. Medical history

The patient is a 28-year-old, monolingual native Dutch-speaking woman from the Netherlands, with monolingual Dutch-speaking parents and an educational level of 13 years. In accordance with the Dutch schooling system, she studied English (for five years), had one year of French and two years of German language instruction. The developmental milestones were timely acquired and scholarly results had always been normal. She worked as an ancillary nurse in a hospital. Her medical antecedents consisted of asthma and migraine. There was no history of alcohol or drug abuse. She had previously experienced two miscarriages and was in the 16th week of a pregnancy when she acutely developed FAS approximately two weeks after a fall down the staircase, when she slid over a loose sheet of paper at the top of the staircase during renovation

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work (June, 2013). The fall did not impact the health of her child. Her relatives and friends perceived her as a non-native speaker of Dutch (initially with a Turkish or Moroccan accent, later Flemish; the variant of Dutch spoken in Belgium).

A clinical neurological examination, laboratory tests and neuroradiological (MRI) investigations were performed after the fall and were normal. A series of psychosocial stress factors were noted: the patient mentioned persistent emotional difficulties as a child after the death of her mother (brain hemorrhage), as well as a very troublesome relationship with her stepmother, which also negatively affected her relationship with her father. She admitted that she had suffered from these issues and that she had been in treatment with a medical psychologist for a suspected post-traumatic stress disorder after her mother's death, although she did not state for how long.

She consulted the department of neurology at ZNA Middelheim in November 2014. The patient complained about headaches, tension, stress, disoriented thoughts and behavior as well as attention difficulties. She stated her accent was now qualified as Belgian Dutch, sometimes French or German. Occasionally, language mixing occurred (German) whenever she was stressed and "could not handle the amount of external stimuli"[28]. The impact of stressors was clinically evident during the intake interview. When researchers and interns entered the consultation room to attend the session, the patient's speech markedly deteriorated and FAS symptoms aggravated. She developed a Flemish-sounding accent, then started to mix German and Dutch words and expressions, e.g. "*Danke, danke sehr*, dokter" ("Thanks, many thanks, doctor").

She claimed that she had no regular contact with Belgian Dutch-speaking people. However, her sister-in-law was German. Following the accent change, she developed loose thoughts and visual hallucinations that lasted for an unspecified amount of weeks. She argued she had equally displayed severely disoriented behavior which also arose for the first time approximately two weeks after the fall: e.g. wanting to go outside through the kitchen window, wanting to take the baby for a stroll at night, observing monkeys in a tree, and at one point she made her suitcase because she "had to leave for Paris" with a stranger she had not seen before. She stated that this strange behavior no longer occurred. These facts were confirmed by the patient's husband. These behaviors were highly egodystonic and provoked an alienating and phrenophobic anxiety.

Upon questioning, she recognized that her emotional problems had been neglected in the past. At the time the patient visited the neurology department she was implicated in litigation.

[28] Patient's own words, during the intake interview in November 3rd, 2014

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8.2.2. Neuropsychological investigations

Table 8.1. Neuropsychological test results November 2014, June 2015, April 2016 and May 2016

Neurocognitive tests	03/11/2014 (raw score or SS) SD or Pc	05/06/2015 (raw score or SS) SD or Pc	24/4/2016 (raw score or SS) SD or Pc	06/05/2016 (raw score or SS) SD or Pc	MEAN (±1SD)
Intelligence					
WAIS-IV-NL					
Verbal comprehension	(83) -1.13 SD				100 (±15)
Perceptual reasoning	(85) - 1 SD				100 (±15)
Working memory	(92) - 0.53 SD				100 (±15)
Processing speed	(84) - 1.07 SD				100 (±15)
Total IQ	(81) - 1.27 S				100 (±1
Memory					
WMS-R					
Attention	(94) -0.4 SD	(87) -0.9 SD		(95) -0.3 SD	100 (±15)
Visual memory	(84) -1.1 SD	(104)+0.3 SD		(109) +0.6 SD	100 (±15)
Verbal memory	(96) -0.3 SD	(76) -1.6 SD		(87) -0.9 SD	100 (±15)
General memory	(91) -0.6 SD	(82) -1.2 SD		(92) -0.5 SD	100 (±15)
Recent memory	(70) -2.0 SD	(76) -1.6 S		(76) -1.6 SD	100 (±15)
RBANS					
Immediate memory	(83) -1.1 SD			(83) -1.1 SD	100 (±15)
Recent memory	(56) -2.9 SD			(52) -3.2 SD	100 (±15)
RAVLT					
Total learning score			(30) pc 1		
Recall			(2) pc 2		
Recognition			(22/30)		
TOMM (memory malingering)			48/50		
Executive functions and attention					
Trail making test A	(29”) pc 70		(36”) pc 14		
Trail making test B	(42”) pc >90		(67”) pc 46		
Stroop test: Card 1	(48”) pc 40		(66”) pc 1		
Stroop test: Card 2	(55”) pc 60		(79”) pc 1		
Stroop test: Card 3	(80”) pc 70		(107”) pc58		
d2-test					
Tn	(393) -1.2 SD			(546) +0.8 SD	
Tn-E	(371) -1.3 SD			(519) -0.79 SD	
CP	(146) -1.5 SD			(211) +0.05 SD	
WCST	(6) +0.38 SD				5.58 (±1.1)
RBANS	(97) -0.2 SD			(88) -0.8 SD	100 (±15)
Verbal fluency	(70) -2.0 SD				60.2 (±21.28)
Language					
Boston Naming Test	(41/60)–3.19 SD	(44/60)-2.4 SD		(28/60) -6.7 SD	52.8 (±3.7)

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Neurocognitive tests	03/11/2014 (raw score or SS) SD or Pc	05/06/2015 (raw score or SS) SD or Pc	24/4/2016 (raw score or SS) SD or Pc	06/05/2016 (raw score or SS) SD or Pc	MEAN (±1SD)
RBANS	(108) +0.5 SD		(101) +0.1 SD		100 (± 15)
Visuospatial skills					
RBANS	(100) ±0.0 SD			(96) -0.3 SD	100 (±15)
Language					
Aachener Aphasia Test (Dutch)					
Comprehension					
Auditory - Words	(24/30)				
Auditory - Sentences	(28/30)				
Reading - Words	(21/30)				
Reading - Sentences	(29/30)				
Token Test	(36/36)				

The first neuropsychological investigation in our department took place approximately seven months after her fall (November 2014, **Table 8.1**). A low average full-scale Intelligence Quotient score of 81 was found (-1.27 Standard Deviation (SD), Wechsler Adult Intelligence Scale-IV-NL (WAIS), Dutch version: Kooij & Dek, 2012). All indices (verbal comprehension, perceptual reasoning, working memory and processing speed index) were in the low average to average range. There was no significant discrepancy between the verbal comprehension (83, -1.13 SD) and perceptual reasoning indices (85, -1 SD). Global attention skills as measured by the Wechsler Memory Scale (WMS-R; Wechsler, 1987) (index = 94; -0.4 SD) and the Repeatable Battery for the Assessment of Neuropsychological Status (Randolph et al., 1998) (index = 97; -0.2 SD) were in the normal range. She scored in the low average to borderline range in sustained visuomotor attention tasks (d2-test: Total number of processed items (Tn): -1.2 SD; Tn- Errors (Tn-E): -1.3 SD, and Concentration Performance (CP): -1.5 SD; Brickenkamp, 1962). Executive functions were normal as reflected by a normal performance on the Wisconsin Card Sorting Test, the Stroop Color-Word Test (Stroop, 1935) (pc. 40/60/70) and the Trail Making Test (Reitan, 1958) (pc. 70/+90). Immediate visual (WMS-R: 84, -1.1 SD) and verbal memory functions (WMS-R: index = 96, -0.3 SD and RBANS: index = 83, -1.1 SD) were within the low average to average range, resulting in an average general immediate memory level (index = 91, -0.6 SD, WMS-R). Delayed memory was impaired: on the WMS-R the patient obtained a deficient index score of 70 (-2 SD). On the RBANS the recent memory index score was profoundly impaired (56, -2.9 SD). Visuospatial skills (RBANS) were normal (100). Performance on the verbal fluency task was also normal (+0.5 SD; unpublished norms). Visual naming was severely impaired (Boston Naming Test (BNT): 41/60, -3.19 SD, Kaplan et al., 1983; Dutch norms: Mariën et al., 1998). Errors predominantly consisted of neologistic (*n*=7) (e.g. 'escalator' - 'wheel chair staircase'), semantic (*n*=7) (e.g.'lion' instead of 'sphinx') and visual (*n*=5) (e.g. 'branch' instead of 'asparagus') mistakes. The Comprehensive Aphasia Test (in Dutch,

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CAT-NL, Visch-Brink et al., 2014) disclosed a weak performance with regard to oral picture description (see Appendix E1), in which grammar and syntax were severely compromised.

Repeat neuropsychological investigations were carried out 7 months later (June 2015). Visual naming was still impaired (BNT: 44/60,-2.44 SD) with errors predominantly of a semantic (*n*=8), visual (*n*=5), and neologistic (*n*=3, e.g. literal: 'hoebel' instead of 'juk' i.e. *yoke*) origin. There was a marked improvement in the immediate visual memory skills (+ 20 index points) (WMS-R: 104, +0.3 SD), while immediate verbal memory functions declined (-20 index points; index 76, -1.6 SD).

In April 2016, The Test of Memory Malingering (TOMM; Tombaugh, 1996) (48/50) performed at the Clinical Centre of Excellence for Body, Mind and Health in Tilburg, The Netherlands, demonstrated that the patient was not malingering or consciously manipulating results. Results on the Stroop test and Trail Making Test regressed slightly and the memory test (Rey Auditory Verbal Learning Test; Rey, 1964) showed marked deficits. In May 2016 cognitive functions were re-evaluated again. Mnestic capacities did not seem to deviate from the results obtained in June 2015. WMS-R performance remained quite stable compared to prior test results, as did the performance on the R-BANS. Performance on the d2-test was more in the average range compared to the results in June 2015. Scores on the BNT however, were now in the severely impaired range (BNT: 28/60; -6.7 SD), these comprised mostly phonological mistakes (*n*=13), neologisms (*n*=6) and visual mistakes (*n*=12), one item could not be named (*n*=1).

8.2.3. Psychodiagnostic examinations

A first psychiatric screening was performed in April, 2014 in a regional hospital in The Netherlands, approximately ten months after the patient's fall (June 2013), because there were indications for psychotic exacerbations. A psychodiagnostic evaluation consisted of the MMPI-2 (Minnesota Multiphasic Personality Inventory- 2, Butcher et al., 1989 - personality test), a shortened version of MMPI in Dutch (NVM; Luteijn & Kok, 1985 - personality test), the Neuroticism-Extraversion-Openness – Five Factor Inventory (NEO-FFI, Costa & McCrae, 1989 - personality test), and the Symptoms Checklist - 90 (Derogatis, 1973). Psychological test results revealed a medium-high score on hysteria scale (MMPI-2). The analysis revealed discomfort in stressful situations. The NVM revealed high scores on the somatization subscale, high score on shyness, and a low score on psychopathology. The below-average (NVM) and average scores (NEO-FFI) on the extraversion, openness scales revealed that the patient could be described as stable, closed, timid and caring. The patient displayed a naive, neurotic personality profile, and had a strong urge for (social) acceptance. There were no indications for psychotic symptoms.

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In December 2014, the patient underwent a second clinical psychiatric assessment. The tests consisted of the MMPI-2 (Butcher et al., 1989), NEO-FFI, the Defense Style Questionnaire-60 (DSQ; Thygesen et al., 2008), the Supernormality Scale (SS; Cima et al., 2003), the Beck Depression Inventory-II (BDI; Beck et al., 1996), the Depressive Experience Questionnaire (DEQ; Desmet et al., 2007), the List of Indeterminate Psychopathology (LIPP; Merten & Stevens, 2012), Rotter Incomplete Sentence Blank (RISB; Rotter et al., 1992), as well as an exhaustive psychological examination (both individually and conjointly with her husband).

Malingering tests (the SS, LIPP and MMPI-2 Validity indices) revealed a slight tendency for a naive positive self-presentation, but without significant effects on test results. No indications for 'fake bad' could be retained. Even on the LIPP she endorsed virtually no pseudo-symptom items. The BDI-2, DSQ-60 & DEQ were presented twice (with a 2 week interval) to check for consistency in the response pattern. Some (randomly selected) items from several tests were discussed, as to check the congruence with anamnestic information, and she could account, contextualize and deepen her answers with insight and emotion.

Her overall personality profile (MMPI, DSQ, DEQ) revealed a sensitive, people-pleasing, dependent personality without any indications for a psychotic structure or symptoms. On the contrary, there were more indications for a stable neurotic personality organization (Kernberg, 1993). The test results could not account for the psychotic behavior, that had been mentioned in past.

Individuals with similar profiles (Within Normal Limit, or 'WNL'-profile with a 3-6-0 pattern) are described as normal individuals, experiencing 'transient situational distress' (Friedman et al., 2014). Their mood is described as stable and happy (as was reflected in a BDI-2 total score of 3 out 63). She no longer described any cognitive complaints herself, though these were formally identified in neurocognitive testing. She described herself as shy, sensitive but globally comfortable in social situations. Although these code-types are associated with past reported transient psychotic symptoms, it remains unclear whether her low scores warrant this association. The questions regarding the etiological nature of the reported psychotic symptoms remained unresolved. The profiles did not suggest a clear potential for somatization or for a conversion disorder. Nevertheless, clear indications were found for a dependent personality. Through analysis of quantitative tests results, projective tests (RISB) and the clinical examination, the neurotic personality organization can be said to be immature. The data largely confirmed the findings of the first psychodiagnostic examination (April 2014), except for the somatization complaints, which could not be confirmed.

Globally, the test results were coherent with one another although quite poor in terms of interpretative content. The face-to-face examination was most informative. During the first interview, her accent was subtle, only slightly noticeable when technical, peripheral or prosaic contents were discussed. But whenever the conversation touched upon affective subjects, her accent and her speech distorted quite rapidly. It remitted back to normal when less emotional issues were discussed. During the inter-

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views, she was agreeable, polite and very open. Although she did not recognize any psychopathology, suffering or mental pain at first, she (through a discussion with her partner) recognized her tendency to occult, repress and split affective experience roughly, and nearly dissociate when negative feelings became too threatening. She appeared benevolently curious in the evaluation and hopeful about its results. When the feedback was provided (the same day) her accent resurged nearly immediately. By changing the topic of the conversation, and the amount of individuals present during the conversation (her partner, staff), thus varying the emotional pressure, cognitive load and fatigue, quantitative distortions in her accent could be induced quite experimentally until a jargon-like speech arose. Although the induction was rapid and nearly one-on-one, its decrease was evident but noticeably slower.

8.2.4. Speech

In addition to the videotaped samples of conversational and spontaneous speech during the consultations, the patient also provided video-taped samples of spontaneous speech which demonstrated accented-ness, language mixing, and neologistic jargon. Samples were analyzed on a segmental and suprasegmental level. In the clinical setting, the patient also performed a structured reading task (text of 231 words) which is analyzed below.

Spontaneous speech

Analysis of the patients' speech samples demonstrated a tendency towards monophthongization, fronting of vowels, and palatalization of consonants, as well as consonant and schwa additions. Dutch, in The Netherlands, is marked by strong tendency towards diphthongization: even monophthongs often tend to be pronounced as diphthongs (Van de Velde et al., 1997; Van de Velde et al., 2010). The patient did not correctly diphthongize most of the diphthongs in speech. Especially the realization of /ɛi/ was markedly different from the expected pronunciation in either variety of Dutch (The Netherlands/Belgium). On the other hand, /e:/ could be expected to be diphthongized as well (/e^{i}/) but the patient only did so on three occasions in the first sample, in word-final position: /o:^{w}ke:^{i}/ (*n*=2/179) and /twe:^{i}/ (*n*=1/179), which is in accordance with the expectations for Dutch (Van de Velde, 1997).

Some words were akin to both German and Dutch (lemma was recognizable) without, however, being correct in either language: e.g. in the second sample (see **table 8.2**), due to the addition of the alveolar fricative /s/ in 'nichts', the word resembled the Dutch word with the same meaning: 'niets' (*nothing*). Despite the addition of a schwa, the word 'kaste'* still closely resembled the Dutch word 'kast' (*closet*). 'Kausen'* resembled the Belgian Dutch (Van Dale, 2016) 'kuisen' (*to clean*). Alternatively 'schunmachen'* is the literal – though faulty – translation of the compounds 'schoon' and 'maken' in the Dutch compound noun 'schoonmaken'. The patient used the same

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strategy when she articulated the word 'schurmacher'* (*sander*), though the compounds are translated wrongfully. In the third sample the phonemic substitution in 'gesag'* (possibly targeting the past participle of the German word 'sagen' - *to say*) which was pronounced as /gɛsɛGt/ (fronting), made it resemble the Dutch 'gezegd'.

In terms of content, the patient produced no neologisms in the first sample of spontaneous speech, though in the second sample she produced four. These were all literal neologisms for which no lemma can be retrieved (see excerpt **table 8.2**): 'mosen*' (*n*=3/45) and 'kanse*' (*n*=1/45). The third sample contained one phonemic neologism that could be transcribed as /noeftze:/.

Although for most words a Dutch or German lemma was recognizable, there equally was interference from other languages. In the second sample the patient uses the phrase 'c'est le ...' which is French for 'it is...', and the word 'televisione', which is Italian for 'television' (see **table 8.2**). In the third sample she uses the word 'exercises', in English.

Table 8.2. Excerpt of the patient's speech containing several literal neologisms (identified with an asterisk):

PATIENT:

"*Sie haben die mosen kaus, die mosen, die mosen da ... ja, und die table, und die televisione ... c'est le kanse die kaste, und andere alles kaste, alles toilette... ja ... alles kausen ... want ich müssen diesen ... alles mit een schurmacher schunmachen ... und mit was ... ja*."

TRANSLATION:

"You have clean the mosen*, the mosen*, the mosen* over there ...yes and the table and the television ... it's the kanse* of the closet, and else everything closet, everything toilet... yes... everything clean ... because I have to this ... clean everything with a sander... and with wax ... yes"

Structured reading task

In November 2014, a structured reading task was performed. This text contains 231 words and is often presented to patients with speech and language disorders (see Appendix E2). The patient made multiple paralexias. In total, her version of the text counted 257 words (excluding filled pauses and interjections). In three instances, she completely changed the word order and occasionally disrupted the syntax:

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1.a. Text: "... kleine groepjes mensen... " [... small groups of people...]
1.b. Patient: *"... kleinen mensengroepen* ..."* [... small people groups*[29]...]
2.a. Text: "... die groepjes vestigden zich ..." [...those small groups settled (down) ...]
2.b. Patient: *"... die die vestigen de groepjes zich[30]* ..."* [... those those settled the groups* ...]
3.a. Text: "... en na verloop van tijd ..." [... and after the course of time ...]
3.b. Patient: *"... en na tijd van verloop* ..."* [... after the time of course ...*]

Phrase 2.b. contains mistakes against Dutch grammar (Subject-Verb-Object structure is not respected). Also remarkable is that phrase 3.b. which is considered as formulaic speech in Dutch, is completely turned around by the patient and rendered nonsensical.

The patient regularly added a schwa to the end of words (irrespective of morphological class) (*n*=12/257): e.g. 'vele' (target: veel= *a lot),* 'geleidelijke' (target: geleidelijk= *gradually*, used as an adverb), 'liepe'* (target: liep= *went*, in text: 3rd pers. sg.), 'tale'* (target: taal= *language*). In other cases, she added an alveolar nasal in the auslaut (/n/) (*n*=4/257). In three instances this created the impression of a declination: she read 'op dezen aarde' instead of 'op deze aarde' (*on this earth/planet*). This may have reinforced the impression of German-like speech, where declination occurs in this position. The pronunciation of the word /ɔntstɔnt/ (developed) was characterized by a postalveolar fricative /ɔnt*ʃ*tɔnt/, a substitution that equally occurred in spontaneous speech.

Suprasegmental characteristics

The speaking rate and articulation rate were calculated with Praat (version 5.4; Boersma & Weenink, 2013). Verhoeven et al. (2004) investigated speaking rate (including silent and filled pauses; Laver, 1994) and articulation rate (excluding silent pauses; Laver, 1994) in a group of unimpaired female native speakers of Dutch (Belgium and The Netherlands) (Verhoeven et al., 2004) and found a mean speaking rate of 4.50 syllables/second (syll/sec) and mean articulation rate of 4.01 syll/sec.

For this patient, the speaking rate for the first spontaneous speech sample amounted to 3.51 syll/sec. whereas the articulation rate amounted to 4.87 syll/sec. In the second sample speaking rate was much slower: 1.58 syll/sec. and articulation rate was 2.98 syll/sec. For the third sample, speaking rate amounted to 1.98 syll/sec. whilst articulation rate was 3.26 syll/sec.

[29] The diminutive in Dutch is marked via a bound morpheme (suffix), whereas in English this requires an adjective. Hence, one to one correspondence in translation is not possible.
[30] "zich vestigen" in Dutch (*to settle*) is a reflexive verb. As this is not the case for the English translation, the reflexive pronoun is not translated.

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In comparison, reading was much more fluent. Van Son & Pols (1992) reported 'reading rates' (number of syllables divided by time in seconds) between 4.4 syll/sec (normal pace) and 6.6 syll/sec (fast pace) in an experienced Dutch speaker based on a structured reading task of 844 words. This seems more or less in line with the values for the FAS patient. The speaking rate and articulation rate were calculated including and excluding syllable repetitions (which only occurred in read speech). Speaking rate was 3.48 syll/sec. and articulation rate was 4.36 syll/sec when including syllable repetitions. When excluding the latter (n=11), speaking rate amounted to 3.83 syll/sec. and articulation rate amounted to 4.80 syll/sec.

In order to qualify rhythm, the Pairwise Variability Index (PVI; Grabe & Low, 2002) was calculated for each of the samples with the help of Praat. The PVI gives an indication of whether a language is stress-timed (e.g. English, Dutch or German) or syllable-timed (e.g. French or Spanish). The normalized vocalic PVI (*nvPVI*) value for Dutch (a stress-timed language) is situated around 65.5. The nvPVI for the first sample amounted to 53.2. The second sample was considerably shorter, instead of calculating PVI with 125 vowels, PVI was calculated based on 69 vowels and the nvPVI amounted to 51.5. For the third sample, the normalized vocalic PVI was calculated based on 106 vowels, and amounted to 50.7. In the classification of Grabe & Low (2002) these values are most comparable to Singapore English (52.3), which is considered a syllable-timed language.

In contrast, the nPVI for the read speech sample amounted to 60.5 (125 vowels), which is indicative of a stress-timed language, although the value is still lower than the value for Dutch (65.5), it corresponds remarkably well to the value Grabe & Low (2002) found for German (59.7).

In contrast to what has often been found for neurogenic (e.g. Graff-Radford et al., 1987; Blumstein et al., 1987; Berthier et al., 1991, case 3; Kurowski et al., 1996) and psychogenic FAS patients (e.g. Gurd et al., 2001; Van Borsel et al., 2005; Verhoeven et al., 2005; Haley et al., 2010; Cottingham & Boone, 2010; Keulen et al., 2016b,c), this patient always placed word stress correctly in spoken speech.

8.3. DISCUSSION

Here FAS is for the first time attested in combination with neologistic jargon. The onset of the accent was related to an episode of intense psychological stress and/or trauma as the patient acquired the accent after a fall down the stairs while pregnant. She herself admitted to have intensely suffered after the death of her own mother when she was a child. In most psychogenic FAS cases, FAS was seen as a (psychologically) stress-induced functional change (see also Charles et al., 2013), possibly co-occurring with a mental disorder, or occurring as a somatic symptom within it (e.g. FAS in conversion disorder). Verhoeven et al. (2005) described a patient who acquired the accent after almost being

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implicated in a car accident. In Keulen et al. (2016c) the patient acquired an accent after being dismissed from her work, supposedly due to a change in temperament some six months after being hit by a car from behind. The accident did not lead to any brain damage. In Reddy et al. (2016) FAS developed after the dissolution of a marriage.

Moreover, our patient had experienced transient psychotic symptoms after the fall. Alienating and phrenophobic anxiety stemming from brief delusional thoughts and visual hallucinations were documented during the interview with the patient herself and the truthfulness of the account was corroborated by her husband. Repeated neurological and neuroradiological investigations formally ruled out the presence of central nervous system damage. Although formal psychodiagnostic assessments did not unambiguously indicate psychopathology, the clinical psychodiagnostic interview showed that touching upon emotional issues triggered changes in the speech (induced a change of accent, and language mixing and switching). Cottingham & Boone (2010) have highlighted the difficulty in objectifying the diagnosis of psychological disorders when the formal psychodiagnostic tests indicate the presence of psychiatric traits without conforming entirely to the criteria of a clearly delineated disorder type in the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, DSM-V, 2013). Their 36-year-old female FAS patient was equally diagnosed with non-credible speech and language disorder(s), including FAS, which was noticed for the first time three years after the motor vehicle accident that the patient held responsible for her symptoms. The MMPI-2 profile did not herald the expected results (a somatoform disorder - conversion profile) and cognitive malingering tests were only borderline significant. The patient displayed behavioral characteristics that were indicative of a psychogenic disorder, which included a left-sided give-way weakness and a feigned deafness. In terms of language, their patient equally displayed an abnormal discrepancy between written and oral output in language testing. The patient displayed deficits in the neurocognitive profile (3 years after a motor vehicle accident), especially pertaining to memory, attention as well as processing speed, even though repeat neuroimaging had formally excluded structural brain damage.

Interestingly, the cognitive profile of the current FAS patient equally demonstrated severe mnestic problems (delayed memory, verbal memory), and relatively intact executive functions and intelligence. In this respect it is also noteworthy that Cottingham & Boone (2010) argue that visual memory tests for malingering (including the TOMM) often fail at disclosing malingering and hence fall short in differentiating, objectively, between credible and non-credible cognitive symptoms (Boone, 2009). Although up until today, clinical experience in conjunction with thorough analysis of the patient's antecedents, cognitive profile, psychiatric traits and behavioral abnormalities is often judged insufficient to corroborate the hypothesis of a psychogenic disorder, it should be clear that the available psychodiagnostic tests often lack the power to allow for an unequivocal diagnosis (see also De Renzi et al., 1997; Keulen et al., 2016b,c; Verhoeven et al., 2005; Van Borsel et al., 2005).

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This patient moreover displayed abnormalities in the linguistic profile. The neurolinguistic tests revealed an abnormal and inexplicable dissociation between the normal results obtained on the tests of the Comprehensive Aphasia Test (CAT; Evy-Visch Brink et al., 2014) and Akense Afasie Test (AAT; Graetz et al., 1992) and deviant results on the BNT (which significantly decreased over the course of approximately one year) and the oral picture description task. This dissociation – even present within the same test session – is inexplicable in the context of neurological damage. Moreover, in terms of spontaneous speech, the fluctuations in accent, language mixing and switching as well as jargon were most remarkable. These symptoms equally seemed to affect the spontaneous speaking more than reading.

Although a few FAS patients have been reported with pathological language mixing and/or switching, the neologistic jargon seems to be an entirely new element in the clinical constellation of FAS. In 2001, Reeves & Norton (see also Reeves & Norton et al., 2007, case 3) reported on a 65-year-old schizophrenic patient who after withdrawal from medication displayed FAS concomitant with psychotic exacerbations. In addition to the change from his original American English accent to a British accent, the patient started using a different code and inserted British English words into his discourse (e.g. 'bloke' instead of 'friend'). The disordered speech in schizophrenics was accounted for as a disturbance of inhibition mechanisms in relation to defective prefrontal functioning, with associated executive dysfunctions (Barr et al., 1989). Language mixing and switching could be explained in the context of a disrupted neurotransmitter balance between prefrontal cortex and basal ganglia (both argued to be essential with respect to the induction of these phenomena). This was resolved when dopaminergic drugs were administered (Moghadam & Jackson, 2004). However, the clearest example of language mixing and switching in the literature on FAS is the 51-year-old female Dutch patient described by Verhoeven et al. (2005). The patient was diagnosed with FAS in the context of a conversion disorder. Her Dutch was characterized by a French accent, a French syntax, morphology and occasionally she used French words or expressions. Even her English was marked by a strong French accent. Importantly, the patient was a teacher of French as a second language, and as such had a good knowledge of the linguistic aspects, not only of the language but also of the interlanguage produced by learners of French when in the process of acquisition. It is the familiarity with the accent and possible faulty usage of lexicon and grammar in second language learners that distinguishes this patient from the current one. Although it is argued that literal translations (a trait that the current patient equally exhibited) could lead to (semantic) neologisms, this was not clear from the examples in the article.

Neologistic jargon as demonstrated by this patient has previously been associated with psychosis and psychopathy (Mazumdar et al., 1991; Kitamura et al., 1995). This patient was not aphasic, so a relation to jargon aphasia was excluded. Most interestingly, FAS could be triggered by conscious manipulation of the subject of conversation. It is not the first time that a link between conversationally related stressors and FAS was attested. Keulen et al. (2016b) reported the case of a native French-speaking patient,

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whose accent was *lost* when an emotional subject was touched upon. In a similar way, fluctuations were psychologically induced.

In our patient, language mixing and the use of neologisms increased when the stressors and/or external stimuli increased. The patient then also started to produce an incomprehensible neologistic jargon. These episodes never remained present for longer than a few minutes, while in aphasic patients, e.g.. those presenting with jargon aphasia, expressive and/or comprehensive deficits persist to some degree (Rohrer et al., 2009).

Segmental and suprasegmental analyses disclosed some of the linguistic characteristics typically associated with psychogenic FAS (Keulen et al., 2016d): a slow speaking rate, fronting of vowels, and additions. However, only in the structured *reading* task the PVI indicated a more German-like speech rhythm, while the patient only used German words in *spontaneous* speech samples. For the spontaneous speech samples however, the PVI index indicated a more syllable-timed rhythm. It seems that the impression of a German accent in spontaneous speech may have been influenced more by segmental aspects and changes at the word level compared to read speech, wherein mainly the prosodic changes were responsible for creating the impression of a German accent.

8.4. CONCLUSION

A case of FAS is described in which – for the first time – neologistic jargon speech was observed. Although an underlying mental disorder could not be formally objectified by psychodiagnostic instruments, the atypical dissociations within the neurolinguistic manifestations strongly indicated a psychogenic etiology. Fluctuations of the speech symptoms could be induced by manipulating stressors. It can be concluded that the neurolinguistic profile and evolution is not consistent with a neurological disorder but with psychogenic FAS.

8.5. ACKNOWLEDGMENTS

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CHAPTER 9

General discussion

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9.1. RESEARCH OBJECTIVES OF THIS DISSERTATION

The general aim of this dissertation was to complement the existing research on Foreign Accent Syndrome by investigating some of the pressing issues that remained unresolved, based on the published literature as well as new case reports. In order to do so, we aimed at achieving the following research objectives:

1) To provide a clear neurolinguistic description of neurogenic, developmental and psychogenic FAS as distinguished by Verhoeven & Mariën (2010a), based on a systematic research synthesis of previously published cases, as well as new case descriptions (Chapters 2-8).

2) Investigate whether FAS is a programming disorder and/or an executive disorder (Chapters 2-4)

3) Investigate the role of the cerebellum in the pathophysiological explanation for FAS (cfr. Cole 1971; Whitaker 1982) (Chapter 2-4)

In this chapter we provide a comprehensive and concise overview of the major conclusions with respect to prior objectives based on the research described in the preceding chapters and we give directions for future research.

9.2. CONCLUSIONS

9.2.1. Neurogenic, psychogenic and developmental FAS are dissociable entities

In Chapter two, we endeavored to review the neurogenic FAS cases and focused on the cases with a vascular etiology (1907 - October 2016). The vascular FAS cases allow for the best lesion-behavior correlations and – usually – a more precise timing of the FAS onset relative to the etiological factor (e.g. stroke, or trauma). We investigated the demographics, lesion location, linguistic features, cognitive profiles, comorbid disorders as well as onset and remission data of vascular FAS. These parameters were also investigated for the psychogenic FAS cases (Chapter 5).

For developmental FAS, we presented an original case study (Chapter 4), although we did not provide a review. The published developmental cases were included in the corpus employed for the selection of vascular FAS cases and the relevant information-could easily be extracted. The amalgam of the published and newly presented cases in chapters 3, 4 and 6-8 permitted an in-depth comparison between the three cognates.

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Demographics

Both vascular FAS (*n*=61) and psychogenic FAS (*n*=21) mainly affected adult, right-handed women based on our samples. Among the vascular FAS patients, 37 out the 61 patients were women, whereas for the psychogenic FAS patients, 15 out of 21 patients were female. However, we cannot extrapolate any conclusion concerning a gender-based distinction in prevalence: the differences were not significant, although nearly so for psychogenic FAS. Developmental FAS mainly affected right-handed men (*n*=4/5). For vascular FAS, we found a mean age of 51 years and 9 months (SD= 12 years, 6 months) and for psychogenic FAS this was 45 years and 11 months (SD= 12 years, 4 months). The developmental population was much younger, with a mean age of 25 years and 2 months (SD= 13 years, 1 week).

Most of the stroke patients in our study were either European or North-American subjects (*n*=44/61). The estimates the World Health Organization (WHO) gives on the prevalence of stroke in Europe (based on data for 28 countries[31]) demonstrate that the prevalence rates for men are usually higher than for women (Truelsen et al., 2006). Our data did not corroborate this finding: there was no gender-related difference in the group of European stroke patients (*n*=26/61) with FAS. On the other hand, studies on stroke prevalence in the USA have shown that prevalence is independent of gender (and situated around 2.5%; Reeves et al., 2008), which is in line with the results of our study when only taking into account the North-American patients (*n*=18/61). However, in the 'Reasons for Geographic and Racial Differences in Stroke'-study (or 'REGARDS' study, United States of America) (Howard et al., 2005) it was shown that prevalence rates for stroke in women and men are quite stable up until the age of 85, when a (double) dissociation suddenly occurs. More (African) American women are diagnosed with stroke (compared to white males and females, and black males) in the age category of 85 and over. In comparison, in the entire study cohort for the investigation of neurogenic FAS (Appendix A1), irrespective of the etiological subtype, only one patient was older than 85 (she suffered vascular dementia).

In line with the findings for psychogenic FAS, women are more prone to mental disorders than men (with the exception of schizophrenia, for which prevalence is not gender-dependent; World Health Organization, 2014). For the developmental speech disorders the prevalence figures are higher in the male than in the female population (Tomblin et al., 1997; Keating et al., 2001; Duffy, 2013).

In terms of linguistic background the results demonstrated that irrespective of the FAS variety, many patients were not monolingual: in the vascular population, it was found that 14 out of 61 patients were bilingual or polyglot speakers (Critchley et al., 1962; Cole, 1971, case 1 & case 2; Schiff et al., 1983; Kurowski et al., 1996; Roth et al.,

[31] These include: Austria, Belgium, Cyprus, Czech Republic, Denmark, Finland, France, Germany, Greece, Iceland, Italy, Luxembourg, Malta, The Netherlands, Norway, Portugal, Spain, Sweden, Switzerland, the UK, Estonia, Latvia, Hungary, Lithuania, Poland, Slovakia, and Slovenia.

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1997; Verhoeven and Mariën, 2002; Laures-Gore et al., 2006, case 1; Karanasios et al., 2011; Levy et al., 2011; Mariën et al., 2013; Moreno-Torres et al., 2013; Verhoeven et al., 2013, case 5; Jezek et al., 2015; and case 2 in Chapter 3); in the psychogenic population the figure amounted to 4 out of 21 patients (Van Borsel et al., 2005; Verhoeven et al., 2005; Haley et al., 2010; Keulen et al., 2016b). In the developmental population 4 out of 5 patients were bilingual speakers (Mariën et al., 2009, Berthier et al., 2016; Keulen et al., 2016a).

FAS onset, evolution and remission

In terms of FAS onset, it was found that vascular FAS often occurs in the acute phase after stroke (*n*=43/61). Psychogenic FAS usually occurred acutely in association with an exacerbation in the context of positive psychiatric symptoms [e.g. mania (Lewis et al., 2013) or psychosis (Reeves & Norton, 2001; Reeves et al., 2007)]. When such symptoms were not present, the delayed time lapse between the incident held responsible for the foreign accent and the FAS onset was often seen as an indicator of a psychogenic etiology (Chapter 5). Although timing was not always provided, we know that for the case of Cottingham & Boone (2010), the patient's accent arose 3 years *after* the motor vehicle accident the patient herself held responsible for the onset of FAS. The patient described in Chapter 7 similarly developed FAS 6 months after being hit by a car, around the time she was dismissed from work (due to a personality change). Interestingly, both these patients were in litigation at the time of investigation. Although it is impossible to unequivocally determine whether this was a (major) influence in the FAS onset or in (suddenly) seeking medical assistance, it is important to take this into account in the diagnostic process and remain vigilant. In the case of the patient described by Cottingham & Boone (2010) the tests assessing malingering[32] were not conclusive, whereas for the cases described in Chapter 7 and 8, malingering was formally ruled out. For developmental FAS, the foreign accent was present as of language development. Hence, FAS onset data seem to be informative in dissociating between the psychogenic cases on the one hand, and the neurogenic subtypes on the other.

The symptom evolution is also instructive with respect to this differentiation. FAS patients with neurogenic and developmental FAS did not show fluctuations with respect to 'accented-ness' – at least not to the point where the foreign accent was completely lost, only to reappear later. This behavior was reported for the psychogenic patients described in Chapters 6-8, as well as the patients who experienced FAS in association with psychotic symptoms (e.g. Reeves et al., 2007).

In terms of remission, 18.03% (*n*=11/61) of the vascular patients remitted (within 3 days to 1 year after follow-up). For the psychogenic patients, FAS receded when exacerbations were pharmacologically inhibited (e.g. Lewis et al., 2013; Reeves and Norton,

[32] Intentional falsifying or exaggerating physical or psychological symptoms for which there is no neurological cause. Motivations to do so can be variable (Rogers, 2012).

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2001; Reeves et al., 2007) or when spontaneous recovery occurred (Van Borsel et al., 2005; Haley et al., 2010). The patients described in Chapters 7 and 8, did not remit during follow-up. The patient described in Chapter 6 suffered FAS and a grammatical output disorder, which receded in oral output while written speech remained agrammatic.

Cognitive deficits associated with FAS

Relatively few vascular FAS patients suffered cognitive deficits. Impairments were noted for only 15.25% of the patients (*n*=10/61) and concerned arithmetic (acalculia), working memory and executive functioning. For the patients reported in Chapter 3, only the first patient had cognitive deficits, which pertained to intelligence, mnestic capacities, executive and initially also frontal functions. The findings were in line with the expectations, as most of these patients suffered damage that affected neural networks subserving (mainly) frontal functions. The lesions were mainly situated within the frontoparietal areas (Nielsen & McKeown, 1961; Kurowski et al., 1996, Scott et al., 2006; Mariën & Verhoeven, 2007, case 2; Naidoo et al. 2008). For PFAS and MFAS1, described by Kuschmann (2010) the lesions were not clearly circumscribed, but situated in 'the left hemisphere'. For the cases described by Mariën et al. (2013), Pyun et al. (2013) and case 1 described in Chapter 3, there was damage to the cerebellum, basal ganglia and pons, respectively. In Chapter 2 and 3 we have argued that FAS is most likely the result of a disruption of the cortico-striato-pallido-thamalic pathways (Schiff et al., 1983; Reeves & Norton, 2001; Blumstein & Kurowski, 2006) or cortico-cerebellar pathways, as the first tract does not include the pons or the cerebellum (and FAS has nevertheless been reported to occur after posterior fossa lesions). It is conceivable that a lesion affecting these tracts, apart from affecting speech and/or language, has an impact on multiple cognitive domains.

The developmental FAS cases obtained satisfying results on cognitive tests, except for the patients described by Berthier et al. (2016) and Keulen et al. (2016a). Cognitive deficits were isolated and different from those reported in the vascular FAS patients. The first patient described by Berthier et al. (2016) demonstrated isolated short-term memory problems (Wechsler Memory Scale - III; Wechsler, 1997), a reduced semantic fluency, and weak scores on the delayed reproduction of the Rey-Osterrieth figure (Rey, 1941). The second patient had isolated deficits on the Hayling test (Burgess & Shallice, 1996), which is a task measuring executive functions, relying on the capacity for response inhibition. Interestingly, the patient obtained normal scores on the Stroop task (Stroop, 1953). This patient had weak performance on the delayed reproduction of the Rey-Osterrieth figure. According to Berthier et al. (2016) the cognitive deficits were largely explainable in the context of the comorbid psychiatric pathology; both patients were diagnosed with obsessive-compulsive disorder, and a prefrontal-striatal dysfunction is supposed in this disorder (Trivedi et al., 2008).

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The patient described by Keulen et al., (2016a) suffered from isolated deficits pertaining to executive functions, visuoconstructive abilities and visuomotor integration. The impairments were explained in the context of defective cerebro-cerebellar networks subserving frontal functions, which exert an important predictive role in sensorimotor control through connections with the parietal lobe, and as such can be related to the comorbid constructive dyspraxia (see also Blakemore et al., 2003).

Cognitive functioning was investigated in 6 out of the 21 psychogenic FAS patients. The impairments were more diverse than in the case of developmental FAS or vascular FAS. However, the etiology was also very disparate. As previously mentioned, the stroke patients with cognitive damage generally had lesions affecting either the cortico-striato-pallidal-thalamic pathway or cerebro-cerebellar pathways, mainly resulting in associated frontal dysfunctions. Psychogenic patients had cognitive deficits affecting memory (Poulin et al., 2007; Jones et al., 2011, Keulen et al., 2016b, case Chapter 8), attention (Poulin et al., 2007; Cottingham & Boone, 2010, Jones et al., 2011; Keulen et al., 2016c), executive functions (Poulin et al., 2007; Cottingham & Boone, 2010; Jones et al., 2011; Keulen et al., 2016c), processing speed (Cottingham & Boone, 2010; Keulen et al., 2016c), intelligence (Jones et al., 2011), fine motor skills (Jones et al., 2011) and visuospatial skills (Keulen et al. 2016c). In most cases these impairments were explicable in the context of the associated psychiatric disorder (Poulin et al., 2007: bipolar disorder; Jones et al., 2011: conversion disorder; Cottingham & Boone, 2010: conversion disorder/cogniform disorder). For the cases described by Keulen et al. (2016b,c), the etiology was less clear, although the case described in Keulen et al. (2016b) was hypothesized to suffer a conversion disorder and had psychiatric traits. Cognitive deficits in Keulen et al. (2016c) were explained in the context of a post-concussion cognitive syndrome. For the patient described in Chapter 8, there were objectified psychiatric symptoms, although the patient did not fulfill the criteria for a psychiatric disorder. The notion of 'cogniform disorder' was introduced to point out the possibility of non-neurologically induced cognitive impairments (Delis & Wetter, 2007).

Comorbid speech and language disorders

In the vascular FAS population (*n*=61) many patients presented with comorbid speech and language disorders. According to our data, 39.34% (*n*=24/61) of the vascular patients were mute in the acute stage before developing FAS. 37.70% of the population developed aphasia (*n*=23/61), among which eight cases also suffered agrammatism. Most of the patients had a non-fluent aphasia (*n*=19/61) and incurred the language disorder after a lesion affecting the precentral gyrus (e.g. Schiff et al., 1983), basal ganglia (e.g. Kurowski et al., 1996), insula (e.g. Verhoeven et al., 2013, case 5) or Broca's area (e.g. Pyun et al., 2013). Fluent aphasia (*n*=2/61) occurred after a lesion affecting the temporoparietal areas (e.g. Kwon & Kim, 2006) and in one case after a lesion affecting the corpus callosum (Hall et al., 2003). Apraxia of speech and dysarthria co-occurred less frequently

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with FAS. According to the data from the review (Chapter 2) as well as the two new case studies (Chapter 3), dysarthria co-occurred with FAS in 27.87% of the vascular FAS cases (*n*=17/61). Both cases reported in Chapter 3 were dysarthric. Apraxia of speech was reported less often, in only 11.48% (*n*=7/61) of the vascular cases. The cases in Chapter 3 did not suffer from apraxia of speech. One explanation for the relatively low diagnostic figures for apraxia of speech can be related to difficulty in differentiating between FAS and apraxia of speech characteristics, especially with respect to segmental changes (see below).

In the developmental cases, FAS was often associated with other developmental speech and language disorders. These comprised developmental apraxia of speech (Mariën et al., 2009, case 1; Keulen et al., 2016a), specific language impairment or SLI (Mariën et al., 2009, case 2), and developmental stuttering (Berthier et al., 2016, case 2).

The comorbid disorders constitute a major point of dissociation between the psychogenic and neurogenic cognate. Compared to the vascular FAS cases for instance, aphasia, apraxia of speech and dysarthria did not occur[33] in the psychogenic population. However, there have been reports of a pre-FAS mutism in psychogenic FAS patients as well as cases of telegraphic speech (Van Borsel et al., 2005, Poulin et al., 2007; Cottingham & Boone, 2010 ; Keulen et al., 2016c,d; Lee et al., 2016). Moreover, in two cases grammar was disrupted due to language mixing and switching (Verhoeven et al., 2005; case Chapter 8). In most instances, the grammatical distortions were argued to be different from classical agrammatism associated with non-fluent aphasia (Van Borsel et al., 2005). One important difference was the specificity and irregularity of mistakes, not always affecting all languages. The latter was illustrated by the cases described by Verhoeven et al. (2005) and the one reported in Chapter 6. Interestingly, for most patients grammatical errors concerned a few, very specific word classes (e.g. Keulen et al., 2016c: only articles and pronouns were affected). Moreover, the problems sometimes waxed and waned (fluctuated) over time (see Chapter 7 and 8), only affected one modality (e.g. Keulen et al., 2016b, agrammatism in written speech and only in the first language, although the patient was a polyglot), and/or were lost due to manipulation of the social context (Keulen et al., 2016b; Chapter 6).

Segmental and suprasegmental characteristics

In **Table 9.1** an overview is provided of the five (perceptual) changes that affected segments (vowels and consonants) and prosody most frequently in the three cognates based on the data presented in the preceding chapters (Chapters 2, 3, 4, 5, 6, 7 and 8).

[33] In Cottingham & Boone (2010) dysarthria and apraxia of speech were reported as non-credible

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Table 9.1 Overview of the five most frequent perceptual changes qualifying speech at the segmental and suprasegmental level in vascular, developmental and psychogenic FAS.

	VASCULAR (*n*=61)	DEVELOPMENTAL (*n*=5)	PSYCHOGENIC (*n*=21)
Segmental			
Vowels			
1	elongations (*n=15*/61)	raising (*n*=3/5)	elongations (*n*=5/21)
2	backing (*n*=10/61)	'substitution' (*n*=1/5)	additions (*n*=5/21)
3	raising (*n*=10/61)	additions (*n*=1/5)	fronting (*n*=4/21)
4	fronting (*n*=8/61)	omissions (*n*=1/5)	monophthongization (*n*=4/21)
5	shortening (*n*=7/61)		lowering (*n*=2/21)
Consonants			
1	manner of articulation (*n*=25/61)	place of articulation (*n*=4/5)	omission (*n*=10/21)
2	place of articulation (*n*=20/61)	omissions (*n*=4/5)	voicing (*n*=7/21)
3	voicing (*n*=14/61)	voicing (*n*=3/5)	manner of arti culation (*n*=6/21)
4	omissions (*n*=10/61)	manner of articulation (*n*=3/5)	place of articulation (*n*=6/21)
5	aspiration is too weak or excessive (*n*=6/61)	additions (*n*=2/5)	addition (*n*=5/21)
Supra-segmental			
1	slow speech rate (*n*=23/61)	speech rhythm (*n*=2/5)	abnormal intonation (*n*=9/21)
2	speech rhythm (*n*=20/61)	slow speech rate (*n*=1/5)	slow speech rate (*n*=7/21)
3	abnormal intonation (*n*=19/61)	high speech rate (*n*=1/5)	speech rhythm (*n*=7/21)
4	wrong word stress (*n*=11/61)	excessive pausing (*n*=1/5)	word stress (*n*=6/21)
5	long pauses (*n*=9/61)		pitch excursions (*n*=4/21)

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Irrespective of the subtype, vowels were more affected by FAS than consonants. It seems difficult to distinguish between the cognates based on purely perceptual phonetic parameters. However, it should be mentioned that narrower linguistic descriptions of FAS cases should be provided in the future in order to allow for more detailed analyses. Nevertheless, based on the data at hand, the previously mentioned dissociations (symptom evolution, onset, associated comorbid disorders) seem to be indispensable in the diagnostic differentiation process. The findings are in line with the arguments of Kurowski et al. (1996) and Ingram et al. (1992): the large set of suprasegmental changes does not suffice to explain the diverse set of segmental distortions. Hence, FAS cannot be seen as a 'purely' prosodic disorder, as argued by Blumstein et al. (1987, *prosodic hypothesis* see Chapter 1). In particular, the consonant changes are difficult to explain in the context of the prosodic hypothesis. The prosodic and segmental alterations can be the result of an advanced tongue root and the associated tense vocal tract setting (Kurowski et al., 1996). The theory of Kurowski et al. (1996) is equally reconcilable with the fortition hypothesis formulated by van der Scheer et al., 2013 (see also Jonkers et al., 2016) as a tense vocal setting would result in more fortis. Problematic with respect to this explanation is that we do not expect contrasting effects to co-occur in the same patient, but this is the case: in psychogenic FAS (Haley et al., 2010) and vascular FAS (Moen, 1990=2006) both fortition and lenition have been observed in the same patient. Hence, although FAS patients often evolve from languages with relatively more reduction and assimilation processes as well as a stress-timed rhythm (which are markers of lenition processes) to languages characterized by less reduction, assimilation, more aspiration, and a syllable-timed rhythm (e.g. English to Spanish or French-sounding, see Jonkers et al., 2016), it is premature to conclude that this mechanism elucidates the speech changes in all FAS cases.

Importantly, these explanations allow for a perception of FAS as an executive *and* as a planning/programming disorder. When interpreting the speech changes as the result of a tense posture for instance, there is a direct relation to dysarthria. Moreover, the fortition hypothesis, as well as the hypothesis formulated by Verhoeven & Mariën (2007) that fortition is a compensatory mechanism increasing feedback (e.g. plosives deliver more feedback than fricatives) can also be interpreted in the context of disrupted speech motor control, as argued by Whiteside & Varley (1998) (see also Varley et al., 2006). This allows for an association with apraxia of speech and brings us to the second point we investigated.

9.2.2. FAS is a dual component disorder

In Chapter 2, we juxtaposed and analyzed the characteristics of vascular FAS, apraxia of speech and ataxic dysarthria. We concluded that FAS should be considered a dual component disorder, based on the shared characteristics with motor speech disorders affecting planning and execution.

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FAS shares an important number of perceptually salient features with apraxia of speech, particularly characteristics of the articulatory type. In terms of the segmental speech dimensions, we found changes of place and manner of articulation in consonants, vowel errors and vowel elongations. At the prosodic level, abnormal intonation and rhythm, slow speech rate and equal stress affect speech in both disorders. Apraxia of speech is also characterized by trial-and-error behavior (groping), which is not frequently seen in FAS patients. Also, in terms of fluency, false articulatory starts as well as syllable repetitions are absent in FAS speakers (Duffy, 2013). Importantly, Darley et al. (1975) argued that apraxia of speech is characterized by more consonant than vowel distortions. The opposite has been regularly argued for FAS as well (Ingram et al., 1992; Miller et al., 2006; Katz et al., 2008; van der Scheer et al., 2013; see also Chapter 2). Perceptually deviant speech features in ataxic dysarthria usually comprise vowel and consonant distortions, and articulatory breakdowns have often been observed (see Chapter 2). Moreover, in contrast to apraxia of speech, it has been argued that vowels are more attained than consonants (Duffy, 2013). At the level of prosody, deviations also correspond quite well to what is seen in FAS patients: altered rhythm, altered stress and slow speech rate qualify speech in both disorders.

Since ataxic dysarthria is an executive disorder and apraxia of speech is a planning disorder, and FAS obviously shares an important number of clinical features with both, the question remained as to where FAS should be characterized: as a planning or as an executive disorder?

In Chapter 2, we referred to the explanation by Scott et al. (2006). They argued that for their patient the disruption between articulatory planning and executive control entailed FAS-like speech. FAS was considered the consequence of a physiological disruption between the motor strip (execution) and anterior insula (planning). Many researchers have come to the conclusion that FAS should be considered a subtype of apraxia of speech on the basis of semiological resemblance of the speech characteristics (Mariën et al., 2009). Some have argued that the disorder should be considered as the mild subtype of apraxia of speech on a continuum on which apraxia of speech would constitute the pole indicating the most impaired status (Varley et al., 2006).

From a neurobiological point of view, it was demonstrated in the second chapter that the majority of the stroke patients had lesions situated either on or near the motor strip (BA 4), the insula and/or the basal ganglia. Ataxic dysarthria is a motor speech disorder that can arise as a consequence of damage affecting the cerebellum or neural circuits of which the cerebellum is an essential part. Even though apraxia of speech is commonly associated with damage to the insula (Dronkers, 1996), there have been cases in which there was a direct association between (right) cerebellar hypoperfusion and apraxic symptoms (Mariën et al., 2001a,b,c). Mariën et al. (2007) have also described remission of FAS symptoms in two patients with supratentorial lesions affecting the left MCA territory (inferior frontal gyrus, precentral gyrus, anterior insula, postcentral gyrus and supramarginal gyrus) (case 1) and the basal ganglia (case 2) after reperfusion of the right cerebellar hemisphere. These cases favor the theory that intact functional com-

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munication between cerebellum and supratentorial speech regions is indispensable to orchestrate (flawless) motor speech production.

9.2.3. The cerebellum subserves a non-negligible function in neurogenic, and possibly also psychogenic FAS

Due to the semiological association between FAS and ataxic dysarthria (see Chapter 2, 3) the question as to the role of the cerebellum in FAS seems particularly relevant. In the current dissertation, the role of the cerebellum was investigated for vascular FAS (Chapter 2), vascular FAS after posterior fossa lesions in particular (Chapter 3), and developmental FAS (Chapter 4).

As mentioned above, from a semiological perspective, the resemblance between symptoms typical of the speech of patients with apraxia of speech and ataxic dysarthria (e.g. difficulty concerning syllable alternate motion rates) is not surprising: the cerebellum communicates with a vast network of cortical regions within the frontal (speech) areas (the supplementary motor area, the premotor and motor area, and the insula). It is evident that loss of excitatory impulses from the cerebellum to these supratentorial areas has important consequences for planning and organization of motor and non-motor linguistic and non-linguistic processes (Mariën et al., 2001a,b,c; Mariën et al., 2006; Chapter 3 and 4).

The cerebellum exerts an important role in predicting movement as well as regulating feedback via the basal ganglia. These functions make it an essential component of motor speech control (e.g. Tourville & Guenther, 2011). As we argued in Chapter 2, the cerebellum plays a significant role in regulating the timing and force of muscular movements to achieve targets, which explains the acoustic outcome in patients with a damaged cerebellum or damage affecting the circuits potentially interrupting cerebellar prefrontal firing (possibly via the basal ganglia, Watson et al., 2014, see also Chapters 3 and 4). As previously argued in Chapter 2, the cortex projects information concerning movement schemes or plans to the cerebellum, which in turn integrates the somatosensory information to make predictions in order to reach targets (its proprioceptive function). Connections with the basal ganglia (especially the thalamus) allow for corrective feedback when required (e.g. Ackermann & Hertrich, 2000; Ackermann, 2008; Boisgontier & Swinnen, 2014).

Importantly, its connection with the basal ganglia via the dentate nucleus allows us to regard the cerebellum as being of major importance in the pathophysiology of FAS: Reeves & Norton (2001), Blumstein & Kurowski (2006) and Schiff et al. (1983) related FAS to a disruption of the cortico-striatopallidal-*thalamic* pathway. This tract subserves many regions that cooperate to regulate different cognitive functions. It has been argued to be involved in various speech and language processes (production and control) (Groenewegen et al., 1990).

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Watson et al. (2014) argued that the effect of cerebellar (and more specifically, dentate nucleus) stimulation on the basal ganglia (esp. substantia nigra and caudate nucleus) can cause a deregulation of the prefrontal dopamine circuits (see also Moreno-Torres et al., 2013). Interestingly, however, in Chapter 5, we argued that modulation of the dopaminergic system can explain psychosis in schizophrenic patients, and was directly related to FAS in the schizophrenic patients described by Reeves & Norton (2001) and Reeves et al. (2007), which potentially reserves a role for the cerebellum in psychogenic FAS. Future research should further investigate the role of neurotransmitter imbalance in the onset of FAS.

9.2.4. Foreign Accent Syndrome ... is a misnomer

When Harry Whitaker coined 'the foreign accent syndrome', he could not foresee the confusion the term would create over the course of the years to come. Whitaker (1982) called the syndrome '*foreign accent* syndrome' because he, and the researchers before him, related the impressionistic phonetic characteristics to very specific foreign (and actually also regional, see Marie, 1907) accents. This, however, when they were relying solely on their ears and their knowledge of foreign languages/accents. Later, phoneticians demonstrated that the ears are bad allies with respect to the recognition of characteristics typical of (known) foreign languages. Often one thinks one recognizes a typical feature of a certain accent, while the particular characteristic is not even produced (e.g. Blumstein et al., 1987; Verhoeven et al., 2013; Berthier et al., 2015; see also the 'foreignness ratings' and 'accent attribution experiments' in Chapters 6 and 7).

Still, even today, the diagnosis of Foreign Accent Syndrome relies completely on the impression it creates in 'the ear of the beholder' (Kurowski et al., 1996). Hence, the saliency of the perceptual features of speech in the hearer is still decisive in the diagnosis of FAS (Miller et al. 2006; Jonkers et al., 2016). However, this is only problematic because a *foreign accent* should be recognized: the physical impression the accent leaves in the hearer is definitely different from the one associated with the native tongue.

Due to the terminology and the Whitaker criteria, other types of accent changes were only hesitantly qualified as FAS, or considered as a disorder within the FAS spectrum: (i) there have been reversions to a previously learned accent (Roth et al., 1997), (ii) there have been reports of a loss of regional accent, where researchers were unable to qualify the 'new' accent because they could not link it to any accent (e.g. see the case of Berthier et al., 2015), and what to do with (iii) patients for whom there was a change of *dialect*? (Kwon & Kim, 2007). The clinicians more likely based their judgment on the fact that a change was perceived in comparison to the premorbid status, rather than recognizing a particular accent.

Another problem is the rejection of the diagnosis of FAS on etiological grounds: in accordance with Whitaker's criteria, the accent change has to be the result of a neurological event. Hence, there is no room for psychogenic or even developmental FAS. We nevertheless have tried to convincingly argue that FAS can be associated with psychiat-

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ric (Chapters 5-8) and neurodevelopmental disorders (see Chapter 4). Furthermore, the criteria leave no room for bilingual and polyglot people. Arguing that a (possibly pathologically induced) accent change in a (non-aphasic) bilingual or polyglot is not an impairment does not hold. Many researchers have identified FAS in bilingual and polyglot patients (see point 9.2.1 above and Chapter 2 and 7).

Deriving a completely different terminology for neurogenic versus other types of FAS on the basis of the suprasegmental and segmental characteristics does not make sense. In the table above (see 9.2.1) the characteristics for neurogenic and psychogenic patients were juxtaposed and there were no real 'between-group' differences, although we have to admit that there was a substantial difference in the number of case reports for both types. Taking into account the inter-individual differences in the symptom constellation, we can only agree with Kurowski et al. (1996), that FAS is "a syndrome, characterized by a constellation of features which may be more or less present and which may be affected in a limited number of different ways" (p. 21). We also conclude that it is safer to dissociate between vascular and psychogenic FAS based on the comorbid speech and language disorders, the absence or presence of psychiatric characteristics, the onset, and the symptom evolution.

In addition, as of chapter 1, the taxonomy presented by Verhoeven & Mariën (2010a) was forwarded as the seminal work classifying different FAS types for the future. However, in 2014 Dilollo and colleagues and in 2016, Schroeder and colleagues described three cases of FAS after a temporo-mandibular-jaw (TMJ) surgery. Although the authors of these papers themselves do not classify their cases, it seems they were at the birth of a new type of FAS. FAS after TMJ surgery in itself does not fit the neurogenic, psychogenic or mixed variant as defined by Verhoeven & Mariën (2010a). In chapter 2, these cases were grouped under a fourth pillar: the organic-mechanic type of FAS. Although the authors of both publications do not focus on the linguistic changes the FAS induces in their patients, future publications should do so.

An accent change induced due to organic-mechanic deficits, and not due to central nervous system damage, will share more characteristics with certain types of dysarthric (executive) disorders than with a planning disorder, such as apraxia of speech. Next to psychogenic and mixed FAS, this fourth FAS type moves the disorder even further from its original conception as a syndrome strictly occurring after central nervous system damage and as such increases the controversy.

Considering that neurogenic FAS only rarely occurs as an isolated phenomenon, and shares many characteristics with apraxia of speech and ataxic dysarthria, doubt as to whether FAS should be seen as a syndrome on its own remains. Even for the psychogenic FAS cases, FAS could be considered as a symptom associated within a psychiatric disorder. Although advances have been made in the understanding of foreign accent syndrome, the debate concerning the status of FAS within the spectrum of motor speech disorders is still ongoing and may be further incited by this dissertation. We hope that by discussing the three main subtypes we have erased some of the reticence to report on new, psychogenic instances of this perceived change in accent. We are opti-

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mistic that the identification of the subtype characteristics in this dissertation will aid the diagnosis of FAS in the clinical setting and by inference will hopefully boost knowledge of the disorder.

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APPENDIX

A. APPENDICES TO CHAPTER 2

Appendix A1. Overview of published Foreign Accent Syndrome case reports. Current table only includes case reports published in national and international peer-reviewed journals, internal medical reports in national and internationally published hospital communications, conference proceedings, abstracts from oral and poster presentations (between 1907 - October 2016).

Abbreviations and test references: 18F-FDG-PET=18F-fluordeoxyglucose-positron emission tomography, ABA-2= Apraxia Battery for Adults-2 (Dabul, 2001); Addenbrooke Cognitive Assessment (Mathuranath et al., 2000), ADL= activities of daily living, AIDS= Assessment of Intelligibility of Dysarthric Speech (Yorkston & Beukelman, 1981); AoS= apraxia of speech, AVM= arteriovenous malformation, AQ= Aphasia Quotient, BA= Brodmann area, BADA= Batteria per l'analisi dei deficit afasici, BDAE= Boston Diagnostic Aphasia Examination (Goodglass et al., 2000), BNT= Boston Naming Test (Kaplan et al., 1983), CLQT= Cognitive Linguistic Quick Test (Helm-Estabrooks, 2003), CT= computed tomography, COWAT= Controlled Oral Word Association Task (Benton et al., 1994), d= day(s), D-KEFS= Delis-Kaplan Executive Function Scale (Delis et al., 2001); DRS= Dementia Rating Scale (Mattis, 1988), DTI= diffusion tensor imaging, EEG= electroencephalogram, F= female, fMRI= functional magnetic resonance imaging, Grooved Pegboard (Ruff & Parker, 1993), h= hour(s), Hayling test (Burgess & Shallice, 1997), IQ= intellectual quotient, K-FAST= Korean version of Frenchay Aphasia Screening Test (Pyun et al., 2009), L= left-handed, l= left, M= male, m= month(s), MAE= Multilingual Aphasia Examination (Benton et al., 1994); MCA= middle cerebral artery, MMPI-2: Minnesota Multiphasic Personality Inventory-2 (Butcher et al., 1989), MMSE= Mini-Mental State Examination (Folstein et al., 1975), MRA= magnetic resonance angiogram, Montréal Cognitive Assessment (Nasreddine et al., 2005), MRI= magnetic resonance imaging, NI= not indicated, NEO-PI-R= Neuroticism, Extraversion, Openness Personality Inventory-Revised (Costa & McCrae, 1992), OCD= Obsessive-Compulsive Disorder, PALPA= Psycholinguistic Assessments of Language Processing in Aphasia PIQ= performance IQ, PVI= pairwise variability index, PVWM= periventricular white matter, R= right-handed, r= right, RAVLT= Rey Auditory Verbal Learning Test (Rey, 1964), RCPM= Raven Colored Progressive Matrices (Raven, 1976; Raven and Court, 1998), SAH= subarachnoid hemorrhage, SCL-90-R= Symptom CheckList-90 Revised (Derogatis, 1992), SPECT= single photon emission computed tomography, Stroop = Stroop Test (Golden et al., 1978), TIA= transient ischemic attack, VIQ= verbal IQ, VOT= voice onset time, TIQ= total IQ, TMT= Trail Making Test (Reitan, 1958), WAB= Western Aphasia Battery (Kertesz, 1982), WM= white matter, WAIS-III= Wechsler Adult Intelligence Scale III (Wechlser 1997), WAIS-R (Wechsler, 1981), Warrington Recognition Memory (Warrington, 1984), WMS-R= Wechsler Memory Scale-Revised (Wechsler, 1987), WRAT= Wide Range of Achievement Test (Wilkinson & Robertson, 2006), y= year(s), Yale-Brown Obsessive-compulsive Scale (Goodman et al., 1989).

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[1] *Marie (1907)	NI/M/NI	Stroke	Left hemisphere involving the lenticular region
[2]* Pick (1919)	26/M/NI	Stroke	Left hemisphere
[3]* Monrad-Krohn (1947)	30/F/NI	Trauma	Left fronto-temporo-parietal
[4]* Nielsen & McKeown (1961) (Case 1)	65/M/R	Thrombosis	Left middle cerebral artery
[5]* Nielsen & McKeown (1961) (Case 2)	38/M/R	Trauma	Left hemisphere contusions
[6]* Critchley (1962) (Case 1)	49/F/NI	Minor trauma -> Neurosis	NI
[7]* Critchley (1962) (Case 2)	48/F/NI	TIA	Left hemisphere
[8]* Critchley (1962) (Case 3)	37/F/R	Stroke	Right hemisphere

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Linguistic Manifestations Type and Course	**Accent**	**Cognitive & Behavioral Symptoms**
at onset: mute for nine years (anarthria); 9 years post-onset: FAS, no aphasia	French Parisian →Alsatian	NI
at onset: mute, aphasia -> fluent, hesitant, paraphasic, agrammatic output, disturbed repetition of unfamiliar names, alexia, agraphia; 4m post-onset: improved aphasia but more prominent FAS	Czech → Polish	Could not sing
at onset: mute, complete aphasia with steady improvement to single words, agrammatism; 7m post-onset: dysphasia, word finding deficits, FAS, intact comprehension; 15m post-onset: improved speech, considerable dysphasia, groping, dysarthria; after 2y: fluent speech, paraphasias, mild agrammatism, paragraphias	Norwegian→German or French-like	Infantile, manic, no emotional control
at onset: mute, motor aphasia, dysarthria, dysprosodia, apraxic agraphia	American English→Swedish	Moderate acalculia
acute: Non-specific dysarthria, grammatical errors	American English→Swedish	IQ above average
NI	English→Welsh	NI
NI	French/English →French (drunken)	NI
at onset: mute, gradual production of single words, good comprehension, agraphia, alexia (dysphasia); 1y post-onset: FAS, adequate vocabulary, fluent speech, impaired writing (no dysarthria, aphasia or AoS)	English → Welsh	Character changes

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[9]* Whitty (1964)	27/M/NI	Bleeding after excision AVM	Craniotomy: AVM in vicinity of the left Rolandic fissure
[10] Critchley (1970) (Case 1) = [6] Critchley (1962)			
[11] Critchley (1970) (Case 2) = [7] Critchley (1962)			
[12] Critchley (1970) (Case 3) = [8] Critchley (1962)			
[13]* Cole (1971) (Case 1)	29/M/NI	Cerebellar anoxia	Clinically suspected cerebellar degeneration secondary to anoxia
[14]* Cole (1971) (Case 2)	58/F/NI	Infarction	Clinically suspected inferior pontine infarction
[15]* Whitaker (1982)	30/F/NI	Stroke	Left hemisphere
[16]* Schiff et al. (1983)	58/M/R	Stroke	CT-scan: lower half of left precentral gyrus with subjacent WM involvement
[17]* Graff-Radford et al. (1986)	56/F/R	Infarction	CT-scan: Brodmann area 6 & WM anterior and superior to the head of the left caudate nucleus

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
at onset: mute for 7 hours; then slow, hesitant and slurred speech; 3d post-onset: normal speech except for FAS and some dysarthric slurring of words, (apraxic?) dysgraphia; 7w post-onset: normal speech, no FAS	English → German	NI
Speech: dysarthric, slightly spastic and dysphonic	Ohian, Yiddish → Italian	No facial apraxia
acute: spastic, dysarthric and dysphonic	Ohian, Hungarian→Eastern European	Normal mental status, no facial apraxia
at onset: muteness evolving to Broca's aphasia with AoS and agrammatism evolving to FAS; some months post-onset: fluent output & FAS	American English→Spanish	NI
at onset: mute; after 7d: mild aphasia; after 3 w: dysarthria, slow speech, dysprosody, FAS, telegrammatic utterances; after 1y: resolution of FAS, permanent dysarthria	Portuguese/English → Chinese	NI
at onset: dysarthria, sparse, labored speech, incomprehensible writing (transcortical motor aphasia); after some days: FAS; 8 m post-onset: FAS and decreased oral word association and slowed reading, lengthened duration of utterances, pauses, inaccurate stresses, sporadic vocal shifts	American English → Nordic	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[18]* Blumstein et al. (1987)	62/F/R	Infarction	CT-scan: 1) deep lesion to left lower sensory-motor cortex area; 2) small subcortical WM lesion deep left middle frontal gyrus and lateral to the putamen; 3) small left temporal gyrus lesion; 4) very small lesion in right middle frontal gyrus
[19]* Ardila et al. (1988)	26/M/R	Infarction	CT-scan: Broca's area (left hemisphere)
[20]* Gurd et al. (1988)	41/F/R	Infarction	CT-scan: lesion extending 1.5 cm lateral to the genu of the left internal capsule involving the lentiform nucleus
[21]* Moen et al. (1990)	61/F/NI	Stroke	Left hemisphere
[22]* Berthier et al. (1991) (Case 1)	70/F/R	Infarction	CT-scan: middle portion of the left precentral gyrus
[23]* Berthier et al. (1991) (Case 2)	58/M/R	Trauma	CT-scan: right fronto-parietal WM (beneath pre- and postcentral gyri)
[24]* Berthier et al. (1991) (Case 3)	47/F/R	Hemorrhagic infarction	CT-scan: lesion involving left middle frontal gyrus

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
acute: sporadic agrammatism in speech, modest word finding difficulties, normal repetition, reading & writing; 9-14d: FAS	American English → Eastern Europe Accent (Slavic to French-like, Dutch of Scandinavian)	VIQ = 84; PIQ = 88, difficulties with complex calculations
at onset: mute with slow recovery of speech over the following weeks; 1m post-onset: telegraphic style; 2m post-onset: FAS, moderate agrammatism (moderate Broca's aphasia)	Spanish →English-like	Normal
at onset: mute -> moaning within hours -> mild dysarthria & FAS (no aphasia); 8m post-onset: residual FAS, diminished verbal fluency	English → French/German	Slight micrographia
acute; FAS (no dysarthria, no aphasia) a few months post-onset: staccato, deviant pitch patterns	Norwegian→English-like	NI
at 48y: 2d mutism & FAS for one month; at 70y: mutism evolving in the acute phase to dysarthria & dysprosodia; 4w post-onset: normal fluency, mild dysarthria, abnormal prosody (FAS) but no aphasia	Spanish→Slavic	Moderate bucco-facial apraxia (initial phase), normal cognition
5d after surgery: mutism no aphasia (crossed aphemia) evolving to dysarthria, dysprosodia; at 2m post-onset: gradual development of FAS; at 1.5y: FAS still present	Spanish→Slavic-like	Normal
at onset: mute & aphasic (Broca's aphasia); at 3w : slow, hesitant speech with impaired articulation, melody and grammar); 1m later: functional communication, writing difficulties, FAS; at 3m post-onset: FAS receded, still dysprosodic	Spanish→Hungarian-like	5m post-onset: normal

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[25]* Berthier et al. (1991) (Case 4)	34/F/R	Trauma	Angiography: right posterior communicating artery & right middle cerebral artery aneurysm CT-scan: posterior superior margin of the left middle frontal gyrus
[26]* Ingram et al. (1992)	56/F/NI	1) Hemorrhage, 2) Trauma	CT-scan: lesion involving left lentiform nucleus
[27]* Seliger et al. (1992)	65/F/L	Infarction	MRI-scan: left centrum semiovale lesion
	44/F/R	Infarction	MRI-scan 4w post-onset: lesion posterior lateral aspect of the left precentral gyrus (middle fifth)
[29] Boatman et al. (1994)	52/M/R	Infarction	CT-scan: lesion of the left superior temporal gyrus extending to the parietal lobe
[30] Denes et al. (1995)	28/F/R	Trauma	CT-scan: left frontal contusion; MRI-scan after 1y: bilateral hypodensities of the frontal WM
[31]* Kurowski et al. (1996)	45/M/R	Infarction	CT & MRI 2y post-onset; 1) left posterior supramarginal gyrus 2) extensive lesion lateral putamen, lateral part of anterior limb of internal capsule, frontal PVWM, anterior & superior to the head of the caudate nucleus

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
at onset post-stroke: mutism for 2d; at day 2: effortful nonfluent speech, speech initiation defects, output limited to perseverations and single words, partial echo-answers, severe agraphia (transcortical motor aphasia); post-surgery (surgery 20 days later): improvement for 2w but speech still dysarthric with staccato cadence & development of FAS; after 3w: receded together with improvement of articulation	Spanish→ Slavic-like	Normal (2m after FAS receded)
at onset: aphasia and AoS; after 1y: FAS and occasional paraphasias	Australian English → Asian, Swedish, German-like	Oral apraxia
at onset: dysarthria and FAS; within 48h: marked improvement of dysarthria; after 2w: moderate accent; after 3w: notably faded accent; at 4m post-onset: almost normal	American English → Northern Irish-like	NI
at onset: mute; after 2d: FAS	Japanese → Korean-like	NI
at onset: expressive aphasia, mild anomia, relatively intact comprehension; within weeks: acute aphasia receded leaving accentedness (dysprosody); after 6m: speech, including prosody within normal limits	American English (?) → Norwegian	NI
at onset: aphonia to dysphonia, dysprosodia, FAS (no aphasia or AoS); 1y post-onset: still FAS	Italian→English-like	NI
at onset: global aphasia; 4m post-onset: mild Broca's aphasia, slight dysarthria; 2y post-onset: normal formal language investigations, normal prosody, FAS	American English → British, Scottish, Irish, Eastern European-like	Slight frontal lobe impairment and impaired digit span; PIQ = 115; normal memory, gnosis, orientation, calculation, oral praxis

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[32] Moonis et al. (1996/1993 abstract)	59/M/R	Trauma	EEG & MRI-scan: normal SPECT 5m post-onset: focal hypoperfusion left inferior dorsolateral frontal lobe & asymmetric response in caudate/putamen perfusion: left < right
[33] Miller & O'Sullivan (1997)	64/F/R	Right anterior aneurysm (junction A1-A2) rupture (SAH)	NI
[34]* Roth et al. (1997)	45/M/NI	Hemorrhagic stroke	CT-scan: left parietal lesion with extension into the ventricles
[35]* Carbary et al. (2000)	51/M/NI	Trauma	CT/MRI: pre-existing infarction of convolutions 2 & 3 of the posterior left frontal lobe
[36] Coelho & Robb (2000)	51/F/NI	Unknown	CT-scan onset: negative MRI-scan (>3d & 1m post onset): normal; EEG: normal
[37] Scianna et al. (2000)	68/F/NI	Infarction	Left parietal lobe
[38]* Dankovičovà et al. (2001)	43/F/R	Infarction	A CT scan: a sub-arachnoid hemorrhage (with evidence of bleeding around the basal system and right Sylvian fissure) and a large terminal carotid aneurysm. After neurosurgery: an extensive infarction in area of right MCA
[39] Gurd et al. (2001)	47/F/R	Multiple Sclerosis?	MRI: bilateral frontal WM, left frontal corona radiata, left thalamus, cerebellar vermis

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
2 & 18m post-onset: abnormal melodic line, mild articulation problems, FAS (no aphasia)	American English→French-like	Normal except for slowed Stroop performance
at onset: stutter-like (no aphasia, dysarthria or AoS)	English → Italian/Polish/ Czech	NI
at onset: mutism evolving to Broca's aphasia and FAS after 2m (no agrammatism or anomia)	American English→Dutch	NI
at onset: mute (catastrophic) with evolution to normal	NI → NI	Normal
(at age 39) at onset: communicated through writing; after 2w: speech returned, left with pronounced accent that cleared after 4y (not formally investigated) (at age 51) at onset: difficulty speaking; after 3d: slow and careful speech with mild word finding difficulties (able to read and spell); after 3w: inconsistent consonant and vowel distortions, WAB quotient=98, intact reading and writing	American English→French Canadian, Italian	Period of confusion Moderate nonverbal oral apraxia
Mild hypokinetic dysarthria & FAS; after 5m: FAS receded	American English → Irish brogue	NI
three weeks post-onset: mild dysarthria	English → Scottish	Impaired nonverbal agility
Transient aphasia (follow-up 16m)	North Yorkshire → French-like	Impaired nonverbal and verbal agility

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[40] Hwang et al. (2001)	40/F/R	Infarction?	MRI at onset: normal SPECT on day one: focal hypoperfusion defect left lateral temporal region and crossed cerebellar diaschisis MRI at 2y: normal SPECT at 2y: normal
[41]* Reeves & Norton (2001)	65/M/NI	Psychosis (Schizophrenia/ Parkinson)	MRI: normal
[42] Simon et al. (2001)	49/F/NI	12/1999: Trauma and Dissection left internal carotid artery 06/2000: brain aneurysm	1999: CT: normal; Repeat CT: after 3d: normal MRI: left internal carotid artery occlusion EEG: moderate right temporal lobe disturbance
[43]* Verhoeven & Mariën (2002)	53/F/R	Infarction	CT/MRI: left hemisphere lesion involving inferior frontal gyrus, precentral gyrus, anterior insula, postcentral gyrus and supramarginal gyrus SPECT: left frontoparietal hypoperfusion extending to left temporal and adjacent subcortical regions, crossed cerebellar diaschisis; SPECT after three years: unaltered supratentorial perfusion pattern, improved perfusion in the right cerebellum
[44]* Hall et al. (2003)	53/F/R	Ischemic Infarction	MRI: body of the corpus callosum

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
No aphasia, dysarthria or AoS but minimal decrease of fluency (by self report), tone errors, FAS; within 3d: almost complete remission of FAS; at 2y : occasional problems with tone in daily conversations	Mandarin → American English-like accent	Normal
No aphasia, dysarthria or AoS	American English → British English	Delusions and hallucinations
at onset: stuttering, mild syntactic errors, mild word retrieval difficulties, FAS ; 09/2000: stuttering, broken speech patterns, sounding Jamaican, adding schwa vowel, normal BNT, normal word fluency, BDAE Cookie Theft picture impaired, prosodic and segmental abnormalities; at 10m post-trauma: significant improvement of fluency and near normalization of prosody	American English → Jamaican/Caribbean accent	Wechsler Memory Scale: normal, normal oral motor function
at onset: verbal mutism for one day evolving to AoS and FAS (no aphasia); 11d –27d post-onset: apraxia of speech and FAS (no dysarthria nor aphasia); 36m post-onset: remission FAS, mild residual apraxia of speech	Dutch → French, Russian-like	Above average cognitive results (IQ, memory, language, praxis, gnosis, concentration, problem solving, executive functions)
at onset: fluent, staccato speech, disturbed melodic line, altered intonation, stresses and pauses, mild fluent aphasia, mild alexia, mild agraphia; after 1y: diminution of staccato speech, written and oral comprehension problems of complex sentences	English → French, French Canadian	No facial or limb apraxia, no tactile anomia

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[45]* Villaverde-González et al. (2003)	38/F/NI	Multiple Sclerosis	CT: two hypointense subcortical lesions in both frontal lobes MRI: several hyperintense lesions at supratentorial (periventricular) and infratentorial level
[46]* Avila et al. (2004)	51/F/R	Trauma	MRI: infarction right temporal lobe and left inferior frontal corona radiata MR angiography: right internal carotid artery obstruction secondary to dissection
[47]* Bakker et al. (2004)	52/F/L	Multiple Sclerosis (for 20 y)	MRI: hyperintense WM lesions (left dorsolateral inferior frontal lobe, corpus callosum, left parietal lobe)
[49]* Coughlan et al. 2004	39/F/NI	Left internal capsule infarction	CT: left internal capsule infarction
[50] Verhoeven & Mariën (2004) = Verhoeven & Mariën (2002) [43]			
[51] Croot et al.(2005) Case 1	13/M/NI	Traumatic brain injury	NI
[52] Croot et al.(2005) Case 2	52/F/NI 52/F/NI	Traumatic brain injury	NI
[53]* Edwards et al. (2005) Case 1	70/F/NI	Traumatic hemorrhage	Left parietal/basal ganglia/ internal capsule
[54]* Edwards et al. (2005) Case 2	58/M/NI	Hemorrhage	Left basal ganglia

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
at onset: slight agrammatism, dysprosody resembling French, decreased speech rate (no dysarthria, no aphasia); after 3m: normalization	Spanish → French	No orofacial apraxia,
at onset: mute with evolution to FAS selectively affecting L1 (Spanish), not L2 (French), L3 (English), L4 (Catalan), no aphasia for more than 2 years	Spanish → NI	NI
at onset: word finding difficulties, minor grammatical errors (no aphasia, dysarthria); four episodes of FAS in 5 years lasting 4 to 6 weeks: slurred and scanned speech, FAS (no dysarthria, apraxia)	Canadian English → Dutch	Decreased memory, labile emotions
Acute: Dysphasia, dysarthria, dysphagia 3 weeks post-onset: amelioration of dysarthria, but onset of FAS: altered rhythm, stress patterns and prosodic features considered atypical for her accent + syllable-timed speech.	(Irish) English → French	NI
at onset: American accent (rhotic vowels and vowel quality) persisted approximately 5 months then disappeared 'overnight' after watching the movie 'Crocodile Dundee'	Australian English → American English	NI
at onset: slurred speech, FAS. FAS diminished but did not resolve.	English → Middle or Eastern European	NI
acute: FAS: receded in 4 days (4 weeks follow-up), normal speech and articulation	English → Welsh	Confusion
acute: Normal speech and articulation, FAS persisted (1 year follow-up)	English → Irish	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[55]* Edwards et al. (2005) Case 3	64/F/NI	Infarction	Left basal ganglia/internal capsule
[56]* Edwards et al. (2005) Case 4	18/M/NI	Trauma	Multiple small left hemisphere contusions
[57]* Edwards et al. (2005) Case 5	53/F/NI	Trauma	Left motor cortex and subcortical contusions
[58] Edwards et al. (2005) Case 6 = Gurd et al. (1988) [20]			
[59]* Fridriksson et al. (2005)	45/M/R	Infarction	MRI: small lesion left putamen MRI tractography: nerve fibers extending from the internal capsule to the corona radiata spared fMRI: normal activations during overt picture naming, increased activity in central sulcus and ventral angular gyrus
[60]* Lippert-Gruener et al. (2005)	35/F/NI	Trauma	CT: traumatic hemorrhage left temporal lobe (cortico-subcortical)
[61]* Munson & Heilman (2005)	49/F/R	Infarction	CT: left middle cerebral artery distribution MRI: left frontal opercular lesion (BA 44)

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
acute: impaired articulation, agrammatism, paraphasias, FAS persisted (9 years follow-up)	Scottish → Dutch/Swedish Russian/German	NI
normal speech and articulation, FAS receded at 3 weeks (3 months follow-up)	English → American	NI
Normal articulation, transient aphasia, agrammatism, paraphasia, FAS persisted (4 years follow-up)	English → Dutch	NI
at onset: severely slurred speech turned into FAS after 2 hours, at six weeks: no aphasia, no dysarthria, no apraxia, normal word finding, diminished but still distinct FAS, fluent speech	American English→French, Greek, British English	Memory, executive functioning: normal, increased sense of smell, taste and appreciation for musical harmony
at onset: amnestic aphasia to severe global aphasia in German (retained capacity to obey commands and to speak a few words in English) evolved to an amnestic aphasia (in German) and dysprosodia, at three months: minor amnestic aphasia and FAS, use of English and Spanish impossible, at six and 12 months: slight improvement, at two years: complete recovery	German → English (at three months)	None at three months
at onset: mute, intact auditory-verbal and written comprehension, normal spelling, after several days: Broca's aphasia and FAS, at one year: fluent speech, mild word finding difficulties, misarticulation of sounds, dysprosody and persisting FAS	Midwestern-type American dialect → German (follow-up of three years)	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[62]* Van Borsel et al.(2005)	32/F/NI	Minor head trauma/ Psychogenic cause	CT: normal
[63]* Verhoeven et al. (2005)	51/F/R	Psychotrauma/ Conversion disorder 1995: conversion disorder (MMPI) 2003: DIS-Q: normal	1995 (onset) CT, EEG: normal 2003 CT, MRI, EEG: normal
[64] Di Dio et al. (2006) = Dankovičová et al. (2001) [38]			
[65] Di Dio et al. (2006) = Gurd et al. (2001) [39]			
[66]* Kwon & Kim (2006)	71/F/R	Stroke	MRI: left temporoparietal infarction MR angiogram: normal
[67]* Laures-Gore et al. (2006) Case 1	64/M/NI	Series of strokes: in 1977, in 1998 and in 1999	1998 MRI: right posterior temporal parietal infarction and an older infarction in the left basal ganglia and left cerebellum 1999 CT: no new infarct
[68] Laures-Gore et al. (2006) Case 2	67/F/NI	?	MRI at age 59: no lesions

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
at onset: mute (by self-report), no aphasia, no verbal apraxia, no dysarthria, deviant articulation, dysprosody, telegraphic style and grammatical anomalies not typical for Broca or Wernicke aphasia, FAS, at one year: resolution of FAS (within 5 months post-onset)	Dutch → Eastern-European, Russian, Slavonic, Romanian, Spanish, French, German, Turkish, non-European	Depression with suicidal ideation
at onset (1995): stutter-like speech and FAS, no aphasia, dysarthria, apraxia of speech, after eight years: no aphasia, apraxia of speech or dysarthria, iterations, effortful speech and inappropriate pauses, FAS originating from lexical, grammatical and pronunciation characteristics, mixing and code switching (FAS in English but not in French)	Dutch → French	Above average cognitive results (IQ, memory, language, praxis, gnosis, concentration, problem solving, executive functions)
20m before: Wernicke aphasia, change of province accent noticed 2d after onset when she started to speak relatively fluent; after 20m: no dysarthria, no AoS, anomic aphasia (WAB=76.5), disturbed reading and writing, affective prosody intact, linguistic prosody impaired	Cholla-buk province accent -> Kwangwon province accent	Slightly depressed
1999: acute aphasia, perceived as FAS; WAB = normal (quotient of 94.8), Apraxia Battery for Adults=mild AoS, no dysarthria	American English → Chinese, →Dutch, or Canadian accent	NI
at age 43: acute change in speech =FAS at age 67: WAB Quotient = 97.5, substitution of 'yes' by 'si', Apraxia Battery for Adults=normal to mild AoS, no dysarthria	American English -> Spanish or Jamaican accent	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[69] Mariën et al. (2006) =Verhoeven & Mariën (2002; 2004) [43,50]			
[70]* Miller et al. (2006) =Miller & O'Sullivan (1997) [33]			
[71] Moen et al. (2006)	End 20s F/NI	MS/psychogenic?	MRI: large subcortical lesion in the right parietal hemisphere. Smaller lesions a.o. near Broca center.
[72] Moen (2006) = Moen et al. (1990) [21]			
[73]* Ryalls & Whiteside (2006)	57/F/NI	Infarction	MRI: small lacunar infarction in left internal capsule
[74]* Scott et al. (2006)	54/F/R	Infarction	MRI: small left hemisphere lesion in WM underneath the precentral gyrus dorsal and medial to the anterior insula
[75]* Varley et al. (2006)	40/F/R	Stroke	MRI: infarction anterior portion left middle cerebral artery territory and hemorrhagic changes left putamen

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
At onset: very serious speech problems, patient had to write to make herself understood. Deviant pronunciation of consonants and vowels. Weak voice, pain during swallowing, normal sensation in face. Duration at least 3 years and 5 months	Norwegian→ Russian	NI
at onset: mute over the following 2m: recovery of speech and development of FAS with 'Briticisms', appropriate intonation, normal BDAE and BNT, no aphasia	American English →British, Australian English	CLQT= criterion level, cognitive functioning within expected age range
at onset: mute, FAS when speech returned; at 2y: fluent speech but sound production and grammatical errors, normal naming, normal comprehension, normal repetition (ignoring the sound production errors), normal writing and spelling	Scottish → German, Polish or South African	At 2 y: WAIS-R: VIQ=94, PIQ=98 ('low' scores for arithmetic and picture completion), verbal recognition memory just above chance level, visual recognition memory slightly below normal limits, normal Weigl sorting test
at onset: nonfluent aphasia, AoS, good resolution of aphasia but FAS; at 2y: mild aphasia, comprehension and naming functionally intact, normal repetition, slow reading and spelling difficulties, occasional errors of morphology and syntax, FAS	South Yorkshire accent → Swedish	Difficulties in divided attention listening situations

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[76]* Luzzi et al. (2007)	64/F/R	Nonfluent Primary Progressive Aphasia	MRI; baseline: normal; after 1y: mild left perisylvian atrophy SPECT after 1y: left frontotemporal hypoperfusion
[77] Mariën & Verhoeven (2007) Case 1 = Verhoeven & Mariën (2002, 2004), Mariën et al. (2006) [69,50,43]			
[78]* Mariën & Verhoeven (2007) Case 2	61/M/R	Hemorrhage	CT: left hemisphere hemorrhage (putamen, genu and posterior limb of internal capsule, extension to the posterior insula, medial temporal lobe and paraventricular white matter of parietal lobe) SPECT: 1m post-onset: severe hypoperfusion left thalamus, lentiform nucleus, left medial and lateral temporal region and left motor cortex, crossed cerebellar diaschisis; 6m post-onset: remission right cerebellar hypoperfusion, unchanged perfusion deficits at the supratentorial level
[79] Moen et al. (2007)	59/M/NI	Infarction	CT: left parietal infarction

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Since about 3y: gradual development of Spanish accent, no aphasia, dysarthria or apraxia; after 1y: hesitant speech, phonological paraphasias, Spanish accent, mild agrammatism, mild to moderate impairments in naming, reading, writing and repetition. Normal oral and written comprehension (mild nonfluent aphasia)	Italian → Spanish	Baseline assessments: normal; after 1y: : no change
at onset: verbal mutism, global aphasia; after 6 days: conduction-like aphasia and FAS; 1m post-onset: conduction aphasia and agrammatism, FAS, very high speech rate, omissions and consonant deletions, harsh voice quality, syllable-timed and isochronous speech rhythm, groping; 6m post-onset: near remission of auditory-verbal and written comprehension deficits, normal naming, residual conduction aphasia, improved speech rate, improved groping and speech rhythm, FAS receded	Dutch →North-African	1m post-onset: disrupted arithmetic
At onset: aphasia and dysarthria After a few months: improved aphasia, mildly agrammatic, word finding difficulties, FAS (no formal indications for AoS), dysarthria (voice problems, reduced phonation and reduced pitch variation, monotonous)	Norwegian → unidentified accent, possibly of Eastern Europe	Reduced psychomotor tempo and sustained attention, memory disorder (storage, recall, working memory), oral apraxia

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[80]* Paquier & Assal (2007)	88/F/R	Binswanger disease (subcortical leukoencephal-opathy- vascular dementia)	CT: profound subcortical vascular leukoencephal-opathy, confluent lesions more pronounced bifrontally (L>R)
[81] Poulin et al. (2007)	74/M/R	?	MRI: asymmetric atrophy left temporal, frontal opercular/insular region (post-hoc) 18F-FDG PET: hypoperfusion bilateral in frontal, parietal and temporal lobes and focal deficit in the left anterior temporal lobe with prominence of the sylvian sulcus
[82]* Reeves et al. (2007) Case 1	30/M/NI	Psychosis (schizophrenia)	MRI; EEG: unremarkable
[83]* Reeves et al. (2007) Case 2	53/F/NI	Psychosis (bipolar disorder)	MRI; EEG: unremarkable

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Recurrent episodes with spontaneous spelling aloud of auditory and visually perceived words and nonwords, FAS, fluent speech, impaired denomination and auditory comprehension, relatively intact semantic knowledge (80%), partially preserved repetition, syntax, orthography, some semantic paragraphias, semantic errors in oral text reading, morphophonological paralexias, omissions of words, tendency to vocalize punctuations marks, normal oral spelling, code switching when speaking French	French→Japanese	Not collaborative, resistant, irritable, agitated, anosognosia, suspicious (no hallucinations or delusions) MMSE=5/30; DRS= 26/144 but not severely demented, basic ADL preserved, partial orientation, utilization behavior, repetitive motor behavior, environmental dependency syndrome
January 2003 at consultation for follow-up exacerbation of bipolar disorder: FAS and agrammatism July 2005: acute exacerbation of bipolar disorder long-lasting FAS and agrammatism; no apraxia, dysarthria, aphasia, German and Spanish sounding nonwords coming to his mind	Québec French→Acadian French, English-like accent	Working memory deficit, executive dysfunctions
at onset: melodic sing-song accent, dropping of consonants, BNT=60, FAS; 6d after olanzapine was resumed: remission of FAS	American English → Jamaican	Disorganized thoughts, bizarre behavior, grandiose delusions; 9d after treatment: improvement
at onset: FAS, rising inflection at the end of phrases, omission of final consonants, no aphasia (normal naming repetition, grammar and comprehension); after 15d of medical treatment: started to lose the accent; at day 17: no accent	American English →European	Elevated mood with racing thoughts, auditory hallucinations, paranoid delusions; 15d after medical treatment: improvement

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[84] Reeves et al. (2007) Case 3= case Reeves & Norton (2001)	?66/M/NI	Psychosis (schizophrenia)	MRI; EEG: unremarkable
[85] Verhoeven & Mariën (2007) = Mariën & Verhoeven (2007) Case 1 = Verhoeven & Mariën (2002, 2004) = Mariën et al. (2006) [43,50,69,77]			
[86] Wendt et al. (2007)	35/F/R	Ischemic Infarction	Left middle cerebral artery infarction at the age of 33
[87]* Hoffmann (2008)	63/F/NI	Cranial trauma (whiplash injury) or viral infection Garcin syndrome	MRI: at three occasions over a follow-up of 2 years = normal. CT: normal Angiogram: normal

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
from 30y onwards: several occasions of FAS during episodes of psychosis; ?: fluent speech, no aphasia or impaired naming or grammar at 66y in nursing home: Episode 1: FAS with incorporation of British terms into vocabulary; elongated vowels, substitution of vowels, substitution of consonants; 4w after medical treatment: resolution of accent; after 2y: Episode 2: FAS with remission after 9d treatment; 6w later: Episode 3: FAS that receded after 9d of treatment	American English → British	Episode 1: hallucinations, delusions, paranoid delusions, agitation; after 4w of medical treatment: persistence of grandiose delusions; after 2y: Episode 2: psychosis with remission after 9d treatment; 6w later: Episode 3: psychosis
2y before current investigation: Broca's aphasia, minor word finding difficulties and mild problems reading aloud, auditory and reading comprehension unimpaired, no alexia, no AoS, no dysarthria, FAS; current examination: problem with rhythmic speech production not characteristic of German or Russian (segmental and prosodic errors)	German → Russian	NI
Onset (1w post-trauma): intermittent voice alteration (French accent), no aphasia (normal BNT); after 3y: remission of accent, residual mild dysarthria with fluent speech and good comprehension	American English→French	MMSE = normal

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[88] Katz et al. (2008)	46/F/R	Unknown	CT and MRI; moderate ventriculomegaly and frontal lobe atrophy
[89]* Naidoo et al. (2008)	50/F/L	Ischemic Stroke	CT: admission: chronic microangiopathic ischemia; day 2: left internal capsule, left basal ganglia and frontal corona radiata lesion, MRI: 1m: anterior & posterior limb of interbal capsule, superior portion of lenticular nucleus
[90]* Tsuruga et al. (2008)	44/F/NI	Dissociative (conversion) disorder	MRI & SPECT; normal

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
2004: slow, measured, dysarthric speech following an allergic reaction to iodine contrast; 2005: complaints of sounding foreign (FAS); lexical substitution; BDAE-3 subtests=normal; BNT 57/60 (diagnosis of mild anomic aphasia); AIDS: single word intelligibility=90%, sentences =96%; ABA-2: mildly impaired diadochokinetic rate, polysyllabic utterance time and repeated trials sub-test; moderate impairment for increasing word length B subtest (mild AoS); fatigue increased accent	American English -> Swedish, Russian, Eastern European	Complaints of short-term memory difficulties and sustained attention to competing stimuli; MMSE=26/30; Trail Making Test=normal; D-KEFS=reduced cognitive flexibility and mild perseverations; normal limb and oral praxis
Onset: slight dysarthria, mild word finding difficulties; 5d poststroke: slow speech rate, imprecise consonants, monopitch, monoloudness, breathy voice quality, word finding difficulties; 14d: no change in speech; 1m: 100% speech legibility at the Assessment of Dysarthric Speech Test, BDAE: impaired recitation skills (1/17), 0 on BDAE melody task, word finding difficulties (BNT 6/15 with circumlocutions, hesitations, semantic paraphasias) changes in phonological segments and expressive prosody; 1.5m poststroke: mild expressive deficits (word finding, expressive vocabulary, confrontation naming, semantic and phonemic verbal fluency); at 3m: greatly improved, word finding difficulties	Native Southern Ontario accent → Canadian East Coast accent (= regional variant)	at 1.5m: right-sided visual inattention, moderate inefficiencies in processing speed, mild reductions in mental control & working memory; mild executive dysfunction
Two transient episodes of aphonia; last period of aphonia evolved into FAS (more significant when talking to family); no aphasia	Japanese→ Chinese	TIQ=101; VIQ=96; PIQ=107; Panic disorder one year after her father's death

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[91]* Abel et al. (2009)	60/F/ R	Breast carcinoma metastasis	CT, MRI: 3 x 3 x 3 cm tumor lesion in the left anterior/inferior parietal lobe (postcentral gyrus), surrounding edema and mild mass effect Intra-operative cortical stimulation mapping (visual object naming): inconsistent hesitations during stimulation of the posterior superior temporal gyrus; no naming areas in the supramarginal gyrus or anterior angular gyrus
[92]* Chanson et al. (2009)	39/F/R	Multiple Sclerosis	MRI: large L prerolandic WM lesion and many small lesions of the deep WM fMRI & DTI tractography: lesion near L corticospinal tract and facial motor area SPECT: R prerolandic, temporal and thalamic diaschisis; after 2m: normal
[93]* Cohen et al. (2009)	58/F/R	Infarction	Stroke 1: MRI: stroke left frontoparietal junction After 3y: Stroke 2: MRI: R inferior cerebellar hemorrhage
[94] *Mariën et al.(2009)	29/F/R	Developmental AoS	CT, MRI & SPECT; normal
[95] *Mariën et al.(2009)	7/M/R	Specific Language Impairment	CT, MRI, EEG; normal SPECT: hypoperfusion in the vermis (-1.96 SD), both lentiform nuclei (left -2.25; right -2.30 SD), left thalamus (-3.30 SD), bilateral occipital lobes

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Onset: dysarthria and accented speech (FAS); immediate postoperative phase: increased dysarthria (communicated with pictures and writing); 3d postoperatively: WAB Auditory comprehension=60%, object identification=100%, reading comprehension=100%, writing=100%; 2w postoperative: dysarthria receded; FAS persisted	American English → Swedish	Unaware of altered speech
German accent (FAS) 2 days after subacute mild right facial palsy (no aphasia, dysarthria, apraxia); within a few weeks: remission FAS	French → German	Normal immediate memory, episodic memory, visuospatial abilities, frontal functions
Onset: aphemia; within hours: English with unlearned accent (FAS) for three years; 2nd Stroke: remission FAS, normal speech	English with unlearned accent	NI
Developmental apraxia of speech (normal oral praxis; aphasia formally excluded)	Native speaker of Dutch perceived with a French-like accent since early childhood	TIQ=88; VIQ=97; PIQ= 81; Verbal MQ=100; Visual MQ=74; normal frontal planning and problem solving; normal visual cognition; normal mood and behavior
Non-fluent speech with phonemic paraphasias, telegrammatic style, agrammatism, normal receptive language skills and written language (SLI: phonological-syntactic syndrome) (normal oral praxis)	Native speaker of Dutch → French-like accent since early childhood	TIQ=124; VIQ=115; PIQ=128; normal visual cognition; normal praxis; normal gnosis; normal problem solving, normal concentration, working memory and verbal memory

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[96]* Teymouri (2009)	53/F/R	Ischemic stroke	CT & MRI: left centrum semiovale lesions
[97]* Cottingham & Boone (2010)	36/F/R	Minor TBI Conversion Disorder (?)	Motor Vehicle Accident CT: (head): normal Headaches 3 days after accident, facial numbness, weakness in R arm, speech difficulties: 10 days after accident Approx. 10 days post-onset: EEG, Brain MRA, MRI: normal, neurological examination: normal, but: AoS + left-sided give-way weakness (non-neurological sign), dysarthria Medical history: complex medical history, with several hospitalizations for symptoms not explicable by neurological cause (e.g. sudden hoarseness of voice)

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Onset: mutism and anarthria, normal reading and auditory comprehension, then FAS?; first m after stroke: gradual improvement of dyslexia and agraphia (weakness in punctuation remained after 2y) a change to a talkative attitude, phonetic, phonemic and morphological difficulties, monotone intonation (variations at 1 y), long pauses between words, wrong stress placement, no sound assimilation	Persian→?	?
Approx. 10 days post-accident: AoS and 'dysarthria' (?); BDAE & Dysarthria Profile: mild retrieval difficulties, hypernasality, halting, abrupt, telegraphic speech (inappropriate pronouns, subject-verb agreement, verb tenses), inconsistent grammatical errors 17-months post-MVA: patient reported reading problems (onset coincided with her 1st deposition in personal injury lawsuit) 22 months post-MVA: Speech characteristics: robotic speech, absent prosody, inconsistent difficulties with articulation, world-list learning task, disfluent speech, omission of articles, prepositions, and plural endings, agrammatism (Cookie Theft)	American English → Eastern accent (3 years after accident)	Behavior: 'overly earnest' and childlike, theatrical presentation. Cognition: (3 years post-onset) Finger tapping, Rey Word recognition + combination, Warrington Recognition Memory, WAIS-III, Rey-Osterrieth, RAVLT Effort, TMT, Stroop, Multilingual Aphasia Examination, BNT, verbal fluency (FAS), MMPI-2 - Impaired scores on WRAT 4 word reading, Stroop Test, MAE sentence repetition, verbal fluency, VIQ was 13 points lower than PIQ - passed for 10/13 indicators of feigned cognitive symptoms

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[98]* Haley et al. (2010)	36/F/NI	Conversion Disorder	MRI: normal MR Angio: normal
[99] Kanjee et al. (2010) = Naidoo et al. (2008) [89]			
[100] Kuschmann (2010) PFAS	61/F/R	2006: Stroke 1 (age 60), at 3w: Stroke 2	Stroke 1: Left hemisphere Stroke 2: ?
[101] Kuschmann (2010) MFAS1	49/F/R	2006: Infarction (age 47)	Left hemisphere
[102] Kuschmann (2010) MFAS2 = Dankovičovà & Hunt (2011)			
[103] Kuschmann (2010) MFAS3	53/M/R	2007: CVA	Left temporo-parietal infarction
[104]* Perkins et al. (2010)	48/F/R	Trauma	MRI (25 days post-onset): abnormal signal intensities in corona radiata + bilateral centrum semiovale EEG: normal
[105] Perkins et al. (2010) = Fridriksson et al. (2005) [59]			
[106] Verhoeven & Mariën (2010a,b) = Verhoeven & Mariën (2007) = Mariën & Verhoeven (2007) Case 1 = Verhoeven & Mariën (2002, 2004) = Mariën et al. (2006) [43,50,69,77,85]			
[107] Akhlagi et al. (2011)	40/M/R	Right temporo-occipital lesion	CT: right temporo-occipital ischemic lesion affecting the calcarine region.

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
onset: slurred speech, abnormal 'cadence'; 14d: fluent in naming and conversation but FAS; subsequent months: frequent relapses heavily to lightly accented, talking like a 6y old child; 4 months post-onset: FAS, formally excluded aphasia (BNT, BDAE, Cognitive Linguistic Quick Test), no dysarthria	American English → French, Spanish, Jamaican, Caribbean, African	NI
onset: slurred speech, no FAS; at 3w (Stroke 2): FAS; following months: less pronounced accent	British English →French, Italian, Eastern European, Jamaican	Stroke 1: short-term memory deficits, acalculia
onset: mutism for a few days; After gradual return of speech: FAS and aphasia	Scottish English → Italian, South African	Severe short-term memory difficulties, dyscalculia
onset: mutism; when speech returned: FAS (aphasia ?, AoS ?)	British English → Italian (as judged by native speakers of English) of English, Polish or Eastern European (as judged by native speakers of Italian)	NI
onset: slurred speech 4 weeks post-onset: FAS	American English→ Eastern European	NI
Onset: mute, when speech recurred: BNT + Token test: only naming deficit, FAS, no dysarthria, no AoS Segmental: especially vowel deficits: backing,	Farsi → Yazdi/ Isfahani	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[108]* Bhandari (2011)	55/M/?	Ischemia	Pre-seizure: CT: encephalomalacia (sign of ischemia) in the left parietal lobe, precentral gyrus and middle frontal gyrus Following seizure: -Transthoracic echocardiogram - EEG - MRI: restricted diffusion in the left parieto-occipital region and in the left middle frontal gyrus, no mass lesion(s) - MRA: normal
[109]* Dankovičovà & Hunt (2011)	56/M/R	2003: Stroke 1 2005: Stroke 2	CT stroke 1: at 6m: normal MRI stroke 1: at 16m: small wedge-shaped area of porencephaly along anterior margin of the mid immediately left to the midline MRI stroke 2 (2008): posterior parietal atrophy superior to occipital sulcus
[110]* Jones et al. (2011)	39/F/R	Conversion Disorder with mixed presentation	CT, MRI: normal EEG: normal

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
raising, centralizing, diphthongization. Consonants: omissions, change of place of articulation Suprasegmental: timing + pitch was altered, stressed initial syllables		
onset: 'foreign' accent, alterations of syllable structure, no change in tone, rate, pitch, no sound substitutions, perseveration, or echolalia, no difficulty with speech initiation, perseveration of syntax, no alexia, no agraphia, was able to copy a paragraph, no acalculia later that day: accent changed back to normal after the seizure, neurological status was otherwise unchanged	Texan English → Cockney English	Mental status: Montréal Cognitive Assessment: 29/30
onset (2003): slurred speech; within a few days: improved speech but emergence FAS (no formal assessments); after 14 month: BDAE: poor non-verbal oral agility task (8/12), normal verbal agility (14/14); stroke 2 (2008): no significant changes of FAS	British English → Italian, Greek (patient grew up in Essex, to Greek mother who spoke English)	Normal except for non-verbal oral agility (BDAE), Apraxia test: transitive limb tasks (9/10), bucco-facial, intransitive limb and whole body tasks (10/10)
onset: mutism; 2-3d: slow rate, articulatory and prosodic abnormalities FAS (no dysarthria or neuromotor deficits); at 20 months: FAS, profound muscular weakness (-4 SD inspiratory and respiratory strength); high-pitched, tremulous and breathy dysphonia in isolated tasks not in connected speech; variable hypernasality; severe right-sided lingual deviation with protrusion; speech sound	American English → Jamaican	At 18m: severe impairment in several domains including memory, executive functions, language, fine motor skills; deficits in general cognition, attention, working memory, auditory comprehension (concerns regarding validity)

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[111]* Karanasios et al. (2011)	76/M/R	Ischemic stroke	CT: on admission: normal; 2d: L temporo-parietal hypodense area MRI: left temporal lesion (posterior superior & middle temporal temporal gyri, inferior supramarginal & angular gyrus & posterior insula) EEG: mild non-specific disturbances over the left temporo-parietal region
[112]* Levy et al. (2011)	42/M/R	Infarction	CT angiogram: left internal carotid dissection CT at 5d: massive left fronto-temporo-parietal lesion; at 3w: more discrete
[113]* Masao et al. (2011)	30/F/NI	Resection of oligoastro-cytoma	CT (2011?): left fronto-temporo- parietal MRI + fMRI (axial T2-weighted) as well as tractography of pyramidal tracts (2011?): confirmation left fronto-temporo- parietal Antecedents: progressively intensifying headaches stretching over the left hemisphere

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
distortions (substitutions, prolongations, omissions, insertions); tongue weakness; multiple prosodic abnormalities; low speech rate; aberrant stress (reduced distinction between stress and unstressed syllables) and intonation (unusual, excess pitch variability)		MMPI: conversion-V profile; NEO-PI-R: very low range on neuroticism scale; SCL-90-R: elevation on the somatization scale (T=65); below 2 SD on STAI
onset: changes in speech expression, FAS, affected prosody (rhythm changes), abnormalities in vowel production (no dysarthria), normal reading & writing, normal comprehension, normal repetition, normal word finding	Origin=Greek, living in Australia for the last 50 yrs. typical Australian-English accent → mixture of Greek and British English accent	Normal visuospatial skills
Onset: nonfluent, agrammatic aphasia without AoS or dysarthria formally tested with BDAE and BAT Follow-up (3 weeks): morpho-syntactic and lexical retrieval deficits, slow speech, hesitations and self-corrections. largely intact comprehension in 3 languages but strong Hebrew accent in English and French Time of the study: residual aphasia with French production most impaired followed by English. Production in Hebrew most preserved	Hebrew (native language) → stronger Hebrew accent in American English (learned at age 10) and French (learned at age 16)	NI
Pre-surgical (neurpsychological) examination: language was coherent and consistent, normal volume, average speed, logical thinking, Spanish native language, left hemisphere dominant for language (DICOTEST). Post-surgery outcome: right hemiparesis and transient changes in language, prosody characteristic of FAS	Spanish (Mexico) → American English	

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[114] Katz et al. (2012) = Katz et al. (2008) [88]			
[115]* Kuschmann et al. (2012) Case 1 = Kuschmann (2010) Case PFAS [100]			
[116]* Kuschmann et al. (2012) Case 2 = Kuschmann (2010) Case MFAS1 [101]			
[117] Kuschmann et al. (2012) Case 3 = Kuschmann (2010) Case MFAS2 = Dankovičovà & Hunt (2011) [102,109]			
[118]* Kuschmann et al. (2012) Case 4 = Kuschmann (2010) Case MFAS3 [103]			
[119] Perera et al. (2012)	30/F/NI	Ischemic infarction	MRI: Ischemic infarction in the left corona radiata and left basal ganglia
[120] Roque et al. (2012)	?/M/?	Ischemic stroke	MRI: ischemic stroke in left pre-motor cortex
[121] Roy et al. (2012) Case 1 = Case Poulin et al. 2007 [81]	75/M/R	(See [81])	(See [81])

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
1 month post-surgery: Expressive language was coherent, logical and normal in its content. Changes in prosody with characteristics of an American English accent, segmental deficits, no aphasia, no dysarthria. 4 months post-surgery: accent persisted		
Acute: vowel distortions, inconsistent consonant conversions and significant deviations in pitch pattern	English → German	NI
FAS, no aphasia, no dysarthria, no AoS Remission after 72 hours after induction of permissive hypertension	British English → Caribbean English	NI
vowels: produced with a restricted F1 and a more open vocal tract position (slightly more open + fronted vowels, esp. for high and mid vowels >< controls), vowel length twice as long as in controls consonants: normal VOT values Suprasegmental char. (text reading): Fo higher than for controls (but still in normal range), speaking rate + articulation rate were slower, PVI lower than for French-speaking controls (French = already considered syllable-timed language with low PVI)	Québec-French speaking → Acadian French (French spoken in the eastern provinces of Canada)	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[122]* Roy et al. (2012) Case 2	63/F/NI	Ischemic stroke	CT: hypodense lesion in the left sylvian area, involving the fronto-parietal cortex
[123]* Lazaro-Perlado et al. (2013)	46/M/R	Hemorrhage	CT: left cerebral hematoma, comprising the striatum and internal capsula CT with contrast: no active bleeding, no structural anomalies in the Circle of Willis, or arterio-venous malformations that can justify a hemorrhage,
[124]* Lewis, et al. (2013)	54/F/NI	Mania	CT (without contrast): during exacerbation: no structural lesions
[125]* Mariën et al. (2013)	71/M/R	Cerebellar stroke (right)	CT: hemorrhagic lesion in right CB, secondary hydrocephalus and bilateral calcifications in the globus pallidus SPECT: a significant decrease and aggravation of perfusion deficits bilaterally distributed in the prefrontal brain regions during follow-up MRI (6 months post-stroke): complete

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
FAS, mild Broca aphasia (mainly characterized by a reduction in spoken fluency) and mixed agraphia	Québec-French → Germanic (German/Alsatian)	NI
onset: FAS, no aphasia 3 years later: remission	Spanish → Gallician	Behavior: OCD with (motor) tics and hoarding behavior (predating FAS and stroke), Cognition: 2 m post-stroke: MMSE: 30/30, Addenbrooke Cognitive Assessment (Spanish): 92/100; Luria test: ok 6 m post-stroke: MMSE: 30/30, Addenbrooke Cognitive Assessment (Spanish): 95/100; Luria test: ok
Fo changes after FAS, condensed vowel space, normal speech rate, excess and equal stress patterns, higher incidence of rising speech contours in statements in FAS, distinctly more melodic line in FAS, substitution errors	North Eastern Carolina American English →Caribbean English	Neuropsychological testing after recovery: no specific diagnostic pattern
onset: FAS (reversion to a previously learned accent) after 10d: akinetic mutism, after resolution: ataxic dysarthria + echolalia + FAS After 1m: spontaneous speech reappeared, FAS and foul language + foreign words (German in his Dutch) Posterior Fossa Syndrome	Dutch → German	1m and 6m post-stroke: depressed MMSE, pathological FSIQ, VIQ and PIQ (WAIS-III), pathological scores on all subtests of the RBANS, pathological scores on WCST, pathological scores on Stroop and TMT` 6m: especially decrease in executive + behavioral test CCAS

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
			resorption of the cerebellar hemorrhage, atrophy of the right cerebellum and restoration of the intraventricular volumes
[126] Mendis et al. (2013)	49/F/NI	Electrocution resulting in brain injury, loss of consciousness	MRI: no abnormalities
[127]* Moreno-Torres et al. (2013)	44/F/R	Stroke (Initially: classified 'psychogenic', diagnosis based in the fact that the initial 1,5 T MRI only disclosed small lesions 'unrelated to symptoms' and the brain PET was normal on visual inspection – analysis with SPM was not performed)	Initial MRI (1,5 T): disclosed three small confluent infarctions involving the left intrasylvian region (frontal operculum + anterior insula) Initial PET: normal Approx. 17 months after first scans: MRI (3 T): small confluenct left hemisphere infarctions located in the inferior frontal gyrus and insula. Largest infarction involved the deep frontal operculum in junction with the ant. insular cortex.

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Language assessment at hospital: in terms of lips; reduced lateral movement in range and speed and reduced speed and range for certain vowels (*u* and *i*), poor coordination of breathing and voice production and poor monitoring of volume throughout the session. The second speech and language session: the patient was taken of beta-blockers and was noted to have a significant increase in disfluent speech, a reduced coordination of breathing and voice, and an increase in coughing. Assessment in a multidisciplinary voice clinic: inappropriately loud, un-coordinated voicing, no obvious spasm. Monotone, flat-pitched voice, and inconsistent volume control. Her speech demonstrated imprecise alveolar placement and some hypernasality.	Northern British English → Polish	Increased anxiety and muscular tension after loss of consciousness
17 months before current examination: R. herpes labialis followed by R. facial paresthesia and weakness involving upper and lower divisions of R. facial nerve Following days: difficulty swallowing food, distinguishing tastes, decreased speech intelligibility during prolonged speaking, poor speech articulation, eventually mute, but could spell (minor spelling errors; letter transpositions): hypothesis of an anterior opercular syndrome (Foix-Chavany-Marie). Auditory + reading comprehension was intact, when anterior opercular syndrome resolved (1 week later) recovery of verbal output was slow (labored, hypophonic, …)	Catalan/Spanish → Czech/ French	17 months after stroke (linguistic analyses in Spanish): WAB (98/100), spontaneous speech was fluent and abundant with normal volume, mild articulation struggling and abnormal rhythm with poor melody. WAB apraxia (60/60). PALPA (nonword minimal pairs= 54/56, word minimal pairs= 55/56, lexical decision= 159/160, word repetition= 23/24, and nonword

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
		Antecedents: In her 20's: Positive history of general lymphaden-opathy due to toxoplasma In her 30's: traumatic cervical injury Since adolescence: recurrent episodes of migraine with aura	DTT: reconstruction suggested reduced intrainsular connectivity an also between areas of infarctions PET: L. hemisphere: significantly decreased metabolic activity in comparison to normal control subjects in cortical and subcortical brain regions implicated in speech processing (inf. frontal gyrus pars orbitalis, dorsal AIC, subgenual cingulated, temporo-polar region, lingual gyrus, putamen, insula, medial/ internal globus pallidus + cerebellum, reduced R. hemisphere metabolism was found involving the medial frontal gyrus, fusiform gyrus, parahippocampal gyrus.
[128]* Paolini et al. (2013)	78/F/R	Primary Progressive Aphasia (nonfluent variant – PPA NFV)	MRI (no date): cortical atrophy, especially in the left temporal hemisphere SPECT: hypoperfusion of lateral frontal regions especially in the left hemisphere, as well as in lateral and medial temporal lobes

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
17 months after stroke (linguistic analyses in Spanish): Further testing: segmental errors (manner of articulation, over-nasalizing of nasals, constriction vowel space esp. as regards F1 >< F2 seemed unaffected, voiced stop consonants → voiceless, fortition of nasal and lateral consonants, fortition of approximants, slow speech rate, intonation is normal, slow articulation rate, linguistic and emotional prosody was disturbed in casual conversation)		repetition= 22/24) and semantic processing (spoken-word picture matching= 40/40) 17 months post-stroke: MMSE (30/30), WMS-III: average performance in word list learning (list A total = 38/48, list B = 6/12, short-term recall of list A = 10/12; recognition = 24/24), digit span score= 8, Rey Osterrieth Complex figure: copy (36/36), delayed reproduction (21.6/36), Trail-Making Test (A = 40s, B = 68.9s), Controlled Oral Word Association Task (45 nouns in 3 min)
03/2008: language disorder characterized by dysprosodia, sporadic phonologic and semantic paraphasias (evolving over 2 years). Symptoms began with progressive change in loudness and pitch of voice, speech was perceived as having undergone a regional accent change. Language evaluation: agrammatic, dysprosodic, anomic spontaneous speech with phonologic errors and rare semantic paraphasias, prosodic comprehension impaired for affective and linguistic prosody, unaware of dysprosody, moderate naming impairment and low phonemic fluency. Normal semantic fluency, single word comprehension was good for nouns and slightly impaired for verbs. Sentence comprehension	Marche accent (central Italy) → Veneto accent (North-east of Italy)	Behavioral changes: disinhibition, impulsiveness, euphoria and irritability Neuropsychological Testing: MMSE: 24/30, follow-up MMSE (12 months later): 9.7, initially a mild frontal executive dysfunction was evident, no deficits in visual and verbal episodic memory, visuospatial abilities and object/people knowledge. Impairment in short-term memory

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[129]* Polak et al. (2013) Case 1	47/M/?	Refractory OCD (for > 25 years) - psychological + pharmacological treatment: no symptom relief	March 2006: 2 DBS electrodes → treatment MRI: pre-operative CT: post-operative No structural lesions (pre- & post-)
[130]* Polak et al. (2013) Case 2	65/M/?	Refractory OCD (> 50 years)	December 2006: 2 DBS electrodes → treatment

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
was severely affected, repetition and writing of non-words were severely impaired, a mild deficit was found for words. Reading ability was good for both words and non-words. 12-month follow-up: severe worsening on oral production and comprehension 24-month follow-up: almost mutatic and oral comprehension was severely impaired, but able to read and to recognize familiar faces.		was found with a low performance in the Digit Span test. Also showed bucco-linguo-facial apraxia 12-month follow-up: a relative preservation of time and space orientation, memory in daily living, visuospatial abilities and selective attention. A mild executive deficit was also confirmed. 24-month follow-up: Her family members confirmed an overall worsening of her behavior with impulsivity, aggression and disinhibition.
Deep Brain Stimulation: DBS after stimulation: speech change: frequently started speaking with an accent commonly used in his native region, accent change co-occurred with hypomanic episode - ! when stimulation parameters were increased to higher voltage: OCD symptoms decreased further, but hypomanic behavior and accent increased, linguistic changes were reported after the adjustments of parameters (until last follow-up in 2011)	Standard Dutch → Regional Dutch accent	- Yale – Brown Obsessive-compulsive Score: decrease from 33 to 18 after stimulation - After cognitive behavioral therapy: from 18 to 5 points
Patient: more coarse language + swearing than before DBS and more hyperactive behavior, characterized by a decrease in the need of sleep and more talkativeness (hypomanic features): decrease in sleep and euphoric mood spontaneously disappeared	Regional variant of Dutch → 'distinguished' Dutch	- Yale – Brown Obsessive-compulsive Score decreased from 33 to 30 after DBS - After cognitive behavioral therapy: further decrease from 30 to 8

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[131]* Pyun et al. (2013)	37/F/	Stroke	Brain CT (admission): intracerebral hemorrhage, left basal ganglia DTI (2 months after stroke): disconnected left arcuate fascicle at the mid-portion adjacent to the hemorraghic lesion. The number of fibers was decreased in the left arcuate fascicle Follow-up DTI: similar morphological features. fMRI*: picture naming task:* in comparison to controls, patient showed different places of activation. More dispersed and extensive. Antecedents: 6y prior to stroke: diagnosed with Moyamoya at the time of her first intracerebral hemorrhage

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
after 8 to 16 weeks. - Increased irritability after approx. 2 to 3 weeks. - After DBS the patient's pronunciation became very distinguished - Hypomanic behavior seemed to be related to adjustment of the parameters, the induced accent remained unaffected by this readjustment - Accent remained present		
One month after stroke: K-FAST (Korean version of Frenchay Aphasia Screening Test)= 16/30, aphasic symptoms showed characteristics of mixed dysarthria + moderate degree of buccofacial apraxia and AoS (inconsistent articulation errors and ineffective self-correction), strange accent (high pitched sounds + elevated the terminal parts of her sentences even when speaking assertively) Speech and language were re-assessed 1m, 6m and 12m after onset: K-WAB initially indicated Broca's aphasia, moderately reduced auditory comprehension, semantic paraphasia and misuse of postpositions in spontaneous speech were observed during reading After speech therapy, AQ evolved from 52 (1 month) to 83.4 (6 months) and 89.4 points (12 months), phonetic analysis of speech, vowels: distortions, prolongation, and diphthongization; for consonants: pattern of final consonant omission and substitution; for suprasegmental changes: prosodic change, unusual rising pitch contours at the end of simple declaratives, prolonged intervals, high fundamental frequency, slow rate, reduced fluency and initial consonant blocking.	Korean →English	One month after stroke: MMSE= 23/30, full cognitive assessment showed moderate to severe impaired performance in attention, memory and executive functions and the impairment was worse in auditory and verbal tasks. Patient received computer assisted cognitive rehabilitation twice a week for a year. Follow-up: 12m after stroke: marked improvement in nonverbal cognitive functions including vigilance and nonverbal intelligence (RCPM). Performance executive functioning worsened.

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[132] Verhoeven et al. (2013) Case 1 = Case [43,50,69,77,85,106]			
[133] Verhoeven et al. (2013) Case 2 = Mariën & Verhoeven (2007), case 2 [78]			
[134] Verhoeven et al. (2013) Case 3 = Verhoeven et al. (2005) [63]			
[135] Verhoeven et al. (2013) Case 4 = Mariën et al. (2009) [94]			
[136]* Verhoeven et al. (2013) Case 5	24/F/R	TIA (2004) Stroke (2005)	CT (2004): normal CT (2004): normal CT, MRI (2005): ischemic lesion in the left insular region SPECT (2006): hypoperfusion in the left motor and insular region (-6.00 SD)
[137] Tailby et al. (2013)	37/F/R	TIA? → Mixed FAS Antecedents: viral encephalitis at 19, pneumonia, gastric banding surgery, ovarian cysts, migraine (one of the migraine attacks involved left-sided weakness and 'loss of sight' in her left eye)	CT of brain, 04/2011 (admission): no acute event CT of brain, 3 days after admission: no acute event MRI (day 12): no acute event, T2 hyperinstensities lateral to anterior horn of left lateral ventricle and in the WM lateral and posterior to atrium of lateral ventricle. DWI (11 days after admission): no hyperintensities fMRI (11/2011): (lexical retrieval paradigm - in-scanner covert version of COWAT + verb generation paradigm): left hemisphere dominant language lateralization (although left-sided weakness), comparison with 32 controls shows that language representation did not significantly differ from that of neurologically normal controls

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
2004: acute onset motor speech symptoms evolving to anarthria within a few hours, normal oral and written comprehension After 12 hours: complete remission of motor speech symptoms (mild residual bradylalia) 2005: sub-global aphasia, evolving within a few days to a mild nonfluent aphasia with agrammatism, AoS and FAS 2006: agrammatism in oral and written language, residual AoS (scanned speech, groping, phonematic errors)	FAS native speaker of English, late bilingual of Dutch (moved to Holland at 8 years of age but had no accent in Dutch) → reversion to a previously learned accent (Birmingham accent affecting Dutch)	2004: verbal apraxia 2006: verbal apraxia, normal memory, normal IQ, normal executive functions, normal attention skills
On admission, 04/2011: slurred speech, 24 hours later (speech pathologist): oro-pharyngeal dysphagia, moderately dyspraxic articulation, slowed and effortful speech, patient spoke of word-finding difficulties 48 hours later (speech pathologist): buccofacial dyspraxia was also noted (/ar/ for /a/) Day 9 of admission: French accent during first fluent conversation (during argument with mother) Following months: speech alternated between at least 5 different accents, could not control the accents and experienced them proprioceptively rather than phonetically, noticed changes in her use of language and pragmatics accompanying each accent 05/2011: no buccofacial or upper limb dyspraxia 08/2011: native Australian accent had returned and patient reported voluntary control over her accent (French/South African)	04/2011: FAS 1: Australian to French, Between 04/2011 – 08/2011: would wake up with either a French or South African or New Zealander accent, 08/2011: Australian to French and South African (controllable!) 18/04-25/04: slow effortful speech 26/04-16/05: French 17/05-20/05: South African 21/05-25/05: French 26/05-02/06: New Zealand accent 03/06-16/06: French 17/06: South African 18/06-19/06: French 20/06-26/06: Russian 27/06-02/07: French 03/07-05/07: French in the morning, Australian in afternoon 06/07-17/07: context driven accent change	Day 4: ataxic gait Between 04/2011 – 08/2011: personality changes congruent with each of the consecutive accents 11/2011: neuropsychological testing: WAIS: VIQ : 85, PIQ: 96, lower ability in language-related cognition is reflected in the WART full scale of 83, new verbal learning and figural recall: within normal limits, processing speed, attention function, working memory and delayed recall were below normative standards.

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[138]* Tomasino et al. (2013)	50/F/R	Low Grade Tumor	MRI: tumor (2.7x2.2x2.5 cm) in left premotor cortex (BA 6, 44, 4) fMRI maps: 6-10d prior to surgery: lip & tongue movements; silent object naming, counting, sentence and pseudoword pronunciation DES: counting & naming
[139]* Tran & Mills (2013)	60/F/NI	Infarction	Initial CT: unremarkable MRI (3 weeks after onset symptoms): subacute infarction of the left hemi-pons and absent flow within the left vertebral artery concerning for occlusion MRA: head+neck re-demonstrated left pontine infarction without hemorrhage, areas of irregular and narrowing in the anterior circulation + diminished flow in the posterior circulation, most pronounced in the basilar artery. MRA with contrast: high-grade stenosis of basilar artery, fetal origin of the left posterior cerebral artery, and multiple areas of narrowing in both MCA's

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
11/2011: reports developing a foreign accent whenever she speaks with someone who has a foreign accent, normal BNT, normal COWAT		
Onset: episodes of focal seizures with speech arrests lasting several minutes; After 1m: onset progressive FAS, reading difficulties and problems linking different parts of speech; Intraoperative mapping: no changes of FAS; Post tumor resection: normal speech, no FAS, no dysarthria, no AoS, no aphasia (formally excluded via Token Test; verbal fluency tasks; BADA phonemic, lexical and syntactic tasks; Robertson Test)	Italian -> Rumanian, Ukrainian/Russian, South African	NI
Initial presentation at emergency dep.: speech change was thought to be secondary to a goiter impinging in the recurrent laryngeal nerve 3 weeks after onset symptoms: presented again to ED, complained of ongoing altered speech pattern On examination: spoke fluently Jamaican / Italian accent, ending most words with '-ah', BNT= 50/60, epenthesis, adding '-uh' to end or middle of words	African-American → Jamaican/Italian	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[140]* van der Scheer et al (2013)	59/M/R	Stroke	MRI: ischemic infarction in left MCA territory and tissue loss in the right occipital lobe consistent with a small, older infarction
[141]* Dilollo et al. (2014) Case 1	46/F/?	TMJ surgery	MRI: inconclusive (artifacts of mandibular reconstruction: metal braces and implants form surgery)

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
After surgery: mute, later his speech improved During neurological examination: no signs of aphasia, only speech rhythm changes, diadochokinesis test was difficult to perform (AoS could not be ruled out, but valid Dutch AoS test was not ready yet), Radboud Dysarthria Test: no dysarthria present Speech and language examination: speech rate is low (2.39 syll/sec), realization of /e/ and /o/ is much more stable than in typically Dutch varieties (realization as [ei] and [ou] respectively)	Dutch → French, German, Asian, but in perceptual experiment: Arabic, Turkish, Surinam, Eastern Dutch accent	NI
Motor speech evaluation: no evidence of diminished strength or mobility of tongue, lips, jaw muscles. Timing and coordination of speech movements were normal, stress testing showed no reduction of accent over time, no speech deviations Acoustic analyses: *vowels*: /i/ in 'heed', /u/ in 'food', /o/ in 'god', /ae/ in 'hat': first + second formants for each of these vowels, in comparison to the allophones in standard American English and British English. Results gave proof of values regarded typical of neither British nor American. Three-year follow-up*:* still suffered from problems due to TMJ surgery (esp. difficulties with pronunciation of multisyll. words). Irish accent was still evident, but less consistently so, able to 'control the accent'	American English (Midwestern English) → (Northern) Irish accent	(Standardized tests not specified): battery that included tests examining visual and auditory learning and memory, executive functioning (planning, switching, word fluency – categorical and phonemic), an effort test, IQ test, a test that estimated pre-morbid IQ, academic achievement tests (spelling, arithmetic, reading comprehension), sensory-motor tasks, language tasks (sentence repetition, auditory and visual confrontational naming, visual receptive naming), personality tests (anxiety, depression, personality traits, psychopathology)

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[142]* Dilollo et al. (2014) Case 2	43/F/?	TMJ surgery	MRI: inconclusive (artifacts of mandibular reconstruction: metal braces and implants form surgery) Three-year Follow-up: migraines→ MRI (hospitalized two times): no structural lesion(s)
[143]* Berthier et al. 2015 (case 1)	47/M/R	stroke	CT: bilateral hemorrhages involving the left motor cortex and right insula-putamen region probably resulting from untreated hypertension or from sporadic cerebral amyloid angiopathy

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
		Results: commensurate with expectations, above-expected up to very superior, no indicators of anxiety or depression were evident
Motor speech evaluation: no evidence of diminished strength or mobility of tongue, lips, jaw muscles. Timing and coordination of speech movements were normal, stress testing showed no reduction of accent over time, no speech deviations Acoustic analyses: *vowels*: /i/ in 'heed', /u/in 'food', /o/ in 'god', /ae/ in 'hat': first + second formants for each of these vowels, in comparison to the allophones in standard American English and British English. Results gave proof of values regarded typical of neither British nor American. Three-year follow-up*:* mild word finding problems	American English (Midwest) → British English	Same as case 1 *Results*: mostly commensurate with expectations, performance on test of receptive vocabulary was almost one SD below expectation, executive functions, attention and concentration: below expectation, no indication of elevated levels of anxiety or depression Three-year Follow-up: no neurocognitive testing, but: patient complained of two episodes (not simultaneous with headaches) where she experienced 'complete memory loss' (now: leaves messages to herself in order not to forget)
Acute: total mutism → verbal mutism (after 2 days) 20 days later: word and sentence comprehension: disturbed, expression and comprehension: normal , writing: disturbed. 8 months post- stroke: agrammatism, echolalia, moderate reading impairment, writing: normal, prosody: different/off, accent change	Cordobés (C-Argentina) Spanish → Italian/? → Argentinian without accent	Depression COWAT: impaired

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
			Neurological examination (8 months post-insult): revealed complete recovery of the right hemiparesis and improvement of language impairment
[144]* Berthier et al. 2015 (case 2)	18/M/R	Cerebral abcess	CT and MRI: encapsulated abscess involving the L sensorimotor cortex with mass effect over the insular cortex and basal ganglia and perilesional oedema Post surgical neurological exam: tongue deviation (right), 'pseudoperipheral' right facial and velum paresis Secondary to an incomplete anterior opercular dysfunction (Foix-Chavany-Marie)
[145]* Jezek et al. (2015)	57/M/	Stroke (Left MCA)	CT: Left MCA infarction, focal lesions in pons
[146]* José et al. (2015)	70/F/NI	Stroke (MCA)	NI
[147] José et al. (2015) = Miller and O'Sullivan (1997) [33] = Miller et al. (2006) [70]	64/F/R	Subarachnoid hemorrhage (right frontal medial)	NI
[148]* José et al. (2015)	47/F/NI	Cerebral vasculitis	NI
[149]* José et al. (2015)	62/F/NI	Stroke (MCA)	NI
[150]* Jose et al. (2015)	37/F/NI	PANDA related action dystonia	NI
[151] Kuschmann & Lowit (2015) – case 1 = Kuschmann 2010 PFAS [100,115]			
[152] Kuschmann & Lowit (2015) – case 2 – Kuschmann 2010 MFAS 1 [101,116]			

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Before surgery: emergency room with a 2-week history of fever, headaches, and vomiting. Upon admission: focal seizures affecting the right face and tongue with secondary generalization and on one occasion transient speech arrest After surgery: mute + aphonic + dysphagia Two weeks after surgery: fluent speech + Japanese accent Flat intonation	Guaranitico (N-E Argentina) Spanish → Japanese? → Argentinian without accent	COWAT: impaired
Initially: mute --> communication through writing After 2-3 hours: difficult articulation --> Russian accent	German→ Russian, Slavic	NI
prosodic, rhythmic/syllable structure alterations and segmental distortions	English → French	NI
Prosodic, rhythmic/syllable structure alterations and segmental distortions	English → Italian	NI
Prosodic, rhythmic/syllable structure alterations and segmental distortions	English → German/Polish	NI
Prosodic, rhythmic/syllable structure alterations and segmental distortions	English → French	NI
Prosodic, rhythmic/syllable structure alterations and segmental distortions	English → Asian	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[153] Kuschmann & Lowit (2015) – case 3 – Kuschmann 2010 MFAS 2 [102,109,117]			
[154] Kuschmann & Lowit (2015) – case 4 – Kuschmann 2010 MFAS 3 [103,118]			
[155]* Liu et al. (2015), case 1	41/F/?	Trauma	CT: left temporal subdural hematoma, contusion in L temporal lobe MRI: confirmation of CT
[156]* Liu et al. (2015), case 2	25/M/?	Trauma	CT: contusion in L temporal lobe and haematoma in dura mater (R hemisphere), diffuse axonal injury. MRI: refused by patient
[157]* Marques (2015)	48/F/R	Dissociative motor disorder (associated with psychiatric problems - marital problems)	MRI: normal EEG: normal Laboratory exams: normal
[158] Reuters (2015), case 1	NI/F/NI	NI	NI
[159] Reuters (2015), case 2	NI/M/NI	NI	NI
[160] Reuters (2015), case 3	NI/F/NI	NI	NI
[161] Reuters (2015), case 4	NI/F/NI	NI	NI

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
NI	Accent lasted 10 days Local dialect → mandarin ?	NI
NI	Accent lasted 1 month Local dialect → mandarin?	NI
NI	Portuguese → German (duration at least 4 months)	NI
Mainly prosodic changes: syllable-timed speech, continuation contours, voicing of typically mute consonants, word finding difficulties	German → French	NI
Segmental level: schwa realized with lip-rounding, absence of glottal-stop before syllable-initial vowels, phoneme omissions suprasegmental level: minimal distinction between stressed/ unstressed syllables → syllable-stressed rhythm, pitch curve that is reminiscent of French	German → French	NI
Segmental: glottalic closure, manner of articulation, wrong word accent, pausing, r-swallowing, vowel lengthening	German → Turkish	NI
Suprasegmental: monotonous, pausing Segmental: backing	German → American English	NI

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[162] Roy et al. (2015)	53/F/NI	? unknown	MRI and CT: normal Diagnosis of PPA was initially made, but later removed
[163]* Sakurai et al. (2015)	42/F/?	stroke	MRI: 4 days post-onset (April 2007): focal infarction surrounding precentral sulcus (upper inf frontal gyrus (BA6) to precentral gyrus at level of lower middle frontal gyrus). SPECT: 19 days post-onset: blood flow reduction in L sup. front. gyrus, post. middle front. gyrus, sensorimotor cortex, ant. and post. supramarg. gyrus, subcortically in the medial globus pallidus. Accent was lost in February 2009, when she suffered 2nd stroke in corona radiata
[164] Tokudo et al. (2015) no translation available			
[165]* Asogwa et al. (2016)	34/F/NI	Schizophrenia	MRI, MRA: normal EEG: normal accent occurred when discharged and less psychotic (!)
[166]* Berthier et al. (2016) case 1	27/M/R	Developmental FAS	MRI: venous angioma close to the head of the L caudate that crossed the medial frontal lobe white matter to drain into the superior sagittal sinus + mild dilatation of the L lateral ventricle DTI: no difference compared to controls, individual differences in

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
fluctuating FAS (very mild to moderate-severe), more present with fatigue; mild anomia; reading difficulties due to attention deficit; Lenition (labial consonants), spirantisation, great variability in formant values, (very mild), agrammatism	Québec French → ? 'foreign accent'	Fluency tasks, Stroop, TMT-A & B, Hayling test, Digit span (forward and backward)
Language evaluation based on WAB (Japanese edition) 12 days post-onset. disfluent spontaneous speech, halting, inconsistent speed, strange pitch accent, and intonation, shortening of word pronunciation, tap consonants (often pronounced as lateral approximant) → pronounced as plosives, agrammatism (Broca's aphasia), vowel shortening, (word) accent shift, vowel + consonant deletion, vowel shift, diphthongization, deviations in VOT compared to healthy controls	Japanese → Chinese/South Korean	NI
Prosodic changes, pitch prominence (words/syllables), decreased pitch range, monotonous speech, consonant changes (change of place, manner, aspect), and changes in vowels (backing of diphthongs)	American English → British English spontaneous reversal of accent at index episode of psychosis/stable(?)	NI
Slight segmental errors, esp. mistakes on phonetic level: consonant strengthening, suprasegmental: ok, although amplitude differences were found in comparison of utterances of FAS with controls	Spanish → French, American English, Argentinean, or Mexican Spanish, Rumanian, and Italian	OCD, depression, general anxiety, social phobia, PTSD, alexithymia, additional personality charateristics: hopelessness, apathy Impaired semantic fluency

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
			some ROI's: Altered DTI-based parameters in the L hemisphere in the superior frontal gyrus, the middle frontal gyrus, and the anterior and posterior cingulate cortex
[167]* Berthier et al. (2016) case 2	46/M/R	Developmental FAS	MRI: expanded perivascular spaces (EPVS) involving both insular cortices DTI: no differences compared to controls, individual differences in some ROI's in the L superior frontal gyrus
[168]* Keulen et al. (2016a)	17/M/R	Developmental Apraxia of Speech	MRI: Normal SPECT: bilateral hypoperfusion distributed in medial prefrontal regions and both lateral temporal regions. Decreased perfusion in the left inferior medial frontal region, right inferior lateral frontal region and right cerebellar hemisphere nearly reached significance.
[169]* Keulen et al. (2016b)	40/F/R	Psychogenic (conversion disorder?)	2005: psychiatric ward (anxiety, aboulia, depressive symptoms) 02/2010: sudden FAS, stuttering, agrammatism, nuchal pains, athralgia, attention problems CT, MRI, EEG: normal 06/2010: Münchausen syndrome/conversion disorder?

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Spatial and motor dysgraphia, fast speech rate (spontaneous speech), Consonant errors included nasalizations, denasalization, place and manner of articulation, vowel insertion, consonant insertion, consonant omission and metathesis	Spanish → Spanish from Lerida (west Catalonia), South-America, Canary Islands, French, or 'uncommon'	OCD, general anxiety, depression, additional personality characteristics: hopelessness, apathy Hayling test: abnormal, Rey - Osterrieth: delayed
Segmental: consonants: voicing errors , uvular –R instead of alveolar -r; vowels: vowel reduction, erosion vowel distinctiveness: esp. /i/, /e/, /Y/. Suprasegmental characteristics: speech rate (SR): 3.83 syll/sec; articulation rate (AR): 4.79 syll/sec = Dutch norms (SR: 3.89 syll/sec, AR: 4.23 syll/sec); Pairwise Variability Index: 48 --> syllable-timed speech, pitch contours (sentences): too many continuation contours (i.e. rising intonation at the end of sentence)	Dutch (Brabantine) → French	Above-average IQ Visuo-constructive competence and visuo-motor integration: problematic Abstract concept formation (executive functions): pathological score on WCST
2010: FAS, agrammatism, stuttering 2011: FAS and stuttering: diminished; agrammatism = still present 2012: FAS and stuttering resolved, agrammatism persisted in writing only FAS characteristics: French uvular-R becomes English diphthong in word final position, addition of –r and schwa, use of voiced velar fricative instead of velar plosive, ejectives, wrong word accent	French → Slavic, Russian, Romanian, Dutch	Verbal memory, logical memory (WMS-R) Rey Complex Figure (< pc. 10)

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
			07/2010: speech problems + gait problems: EEG: normal, MRI: normal 04/2011: psychiatric ward admission CT + EEG: normal agrammatism 08/2012: resolution of FAS after general anesthesia for appendectomy, agrammatism in writing only
[170]* Keulen et al. (2016c)	33/F/R	Psychogenic: bordeline personality	12/2011: minor head trauma; CT: normal 1 week later: repeat CT + EEG: normal 11/2012: MRI: normal
[171]* Lee et al. (2016)	59/F/NI	Psychogenic FAS - post-traumatic stress disorder	Facial trauma (tripped): lost front teeth CT: normal MRI: area of right occipital white matter high signal; this was thought to be consistent with a perinatal event and not relevant either to the head injury or the current presentation Dental surgery (repair teeth after accident): accent change soon after anesthesia for dental surgery

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
No aphasia, no comorbid disorders but isolated morphological errors (articles) FAS characterized by slow speech rate, subtle segmental changes, altered rhythm (PVI) Fluctuating accent	French → Dutch	2012: WAIS: dissociation between verbal IQ (96) and performance IQ (120) Stroop: slow processing speed d2: slow processing + accuracy : pathological range 2014: WMS-R: dissociation between verbal memory index (74) and visual memory index (133), general attention index was in clinical range (70)
Occasional segmental errors (manner and place), speech sometimes sounded telegraphic, used German sounding words in her speech	Scottish English → Scandinavian, German, fluctuating accent 'German-like' - Scottish (reversal noted on two occasions), patient could also imitate posh English and Irish accent, after therapy: periods of remission interspersed with accented speech Accent 'reinforced' sense of altered identity	Addenbrooke' s Cognitive Assessment: normal sense of altered identity post-traumatic stress disorder

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Case Reference	Age / Gender/ Handedness	Etiology	Lesion Location
[172] Schroeder et al. (2016)	42/F/A or R?	possible CNS lesion induced by TMJ surgery (born with prominent mandibular prognathism, TMJ pain as of age 14)	TMJ: oral and maxillofacial surgery was performed at age 42 CT, MRI, EEG (4 months after surgery/onset accent): normal

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Linguistic Manifestations Type and Course	Accent	Cognitive & Behavioral Symptoms
Acute (post-surgery): word-finding problems, British accent, difficulty with reading	American English! → British accent	Post-surgical emotional change (no depression, no anxiety), feelings of raised irritability Significant dissociation between general memory (117) score and WM (85) ($p < 0.05$), verbal fluency (-2 SD), WM (-1.9 SD), letter-number sequencing (-1.9 SD), digit span (-2.2 SD), Trail Making Test (switching: -2.2 SD), Verbal fluency (letters): -2.9 SD, Grooved Pegboard Right Hand (-2.1 SD) (but: prior right radial nerve decompression surgery, date unknown)

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B. APPENDICES TO CHAPTER 3

Appendix B1. FAS after posterior fossa lesions.

Abbreviations: Ant.=anterior, BDAE= Boston Diagnostic Aphasia Examination (Goodglass et al., 2000), BNT= Boston Naming Test (Kaplan et al., 1983), CT= Computed Tomography, ED= emergency department; F=female, inf.= inferior, L= left, m= month(s), M=male, MCA= Middle Cerebral Artery, MRA= Magnetic Resonance Angiography, MRI= Magnetic Resonance Imaging; NI= not indicated, post.= posterior, R= right, R*= right-handed, SPECT= Single Photon Emission Computed Tomography, sup.= superior, w= weeks(s); y= year(s).

Case	**Age/ gender/ handedness**	**Etiology**	**Lesion Location**
Case 1: Cole (1971) (Case 2)	29/M/NI polyglot	Cerebellar anoxia	Clinically suspected cerebellar degeneration secondary to anoxia
Case 2: Cole (1971) (Case 1)	58/F/NI polyglot	Infarction	Clinically suspected inf. pontine infarction
Case 3: Dankovičovà & Hunt (2011)	56/M/R* monolingual	2003: Stroke 1 2005: Stroke 2	Stroke 1: CT at 6m: normal MRI at 16m: small wedge-shaped area of porencephaly along anterior margin of the mid pons immediately to the midline MRI (2008): post. parietal atrophy sup. to occipital sulcus
Case 4: Tran & Mills (2013)	60/F/NI	Infarction	Initial CT: normal MRI (3w post-onset): subacute infarction in the L hemi-pons and absent flow within L vertebral artery MRA head and neck: left pontine infarction (no hemorrhage), irreg-

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Speech/language characteristics	Original accent	Newly acquired accent	Cognition
Acute: dysarthric, slightly spastic and dysphonic	Ohian, Yiddish	Italian	No oral apraxia
Acute: spastic dysarthric and dysphonic	'Ohian' (i.e. American English) and Hungarian (polyglot!)	Eastern European	Normal mental status, No facial apraxia
Onset (2003): slurred speech; Within a few days: improved speech but onset FAS (no formal assessments) After 14m: BDAE: poor non-verbal oral agility task (8/12), normal verbal agility (14/14) Stroke 2 (2005): no significant change of FAS	British English	Italian, Greek (patient grew up in Essex, to Greek mother who spoke English)	Normal except for non-verbal oral agility (BDAE), apraxia test: transitive limb tasks (9/10), bucco-facial, intransitive limb and whole body tasks (10/10)
Initial presentation at emergency department: speech change was thought to be secondary to a goiter impinging in the recurrent laryngeal nerve 3w after onset symptoms: presented again to ED, com-	African-American	Jamaican/ Italian	NI

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Case	Age/ gender/ handedness	Etiology	Lesion Location
			ularity in ant. circulation, diminished flow in the post. circulation (affecting esp. basilar artery) MRA with contrast: high-grade stenosis of basilar artery, fetal origin of the L posterior cerebral artery, and multiple areas of narrowing in both MCA's
Case 5: Cohen et al. (2009)	58/F/R*	Infarction	Stroke 1: MRI: stroke L fronto-parietal junction After 3y: stroke 2: MRI: R inf. cerebellar hemorrhage
Case 6: Mariën et al. (2013)	71/M/R	Cerebellar stroke (right)	CT: hemorrhagic lesion in right CB, secondary hydrocephalus and bilateral calcifications in the globus pallidus SPECT: a significant decrease and aggravation of perfusion deficits bilaterally distributed in the prefrontal brain regions during follow-up MRI (6 months post-stroke): complete resorption of the cerebellar hemorrhage, atrophy of the right cerebellum and restoration of the intraventricular volumes

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Speech/language characteristics	Original accent	Newly acquired accent	Cognition
plained of on-going altered speech pattern: spoke fluently Jamaican / Italian accent *ending most words with '-ah', *epenthesis: adding schwa to end or middle of words BNT= 50/60,			
Onset aphemia; within hours: English with unlearned accent (FAS) for three years Stroke 2: remission FAS, normal speech	English	English with un-learned accent	NI
Onset: FAS (reversion to a previously learned accent) After 10d: akinetic mutism, after resolution: ataxic dysarthria + echolalia + FAS After 1m: spontaneous speech reappeared, FAS and foul language + foreign words (German in his Dutch) Posterior Fossa Syndrome	Dutch	German	1m and 6m post-stroke: depressed MMSE, pathological FSIQ, VIQ and PIQ (WAIS-III), pathological scores on all subtests of the RBANS, pathological scores on WCST, pathological scores on Stroop and TMT 6m: especially decrease in executive + behavioral test CCAS

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Appendix B2. FAS after frontal lesions with related functional involvement of posterior fossa structures (diaschisis).

Abbreviations: AAT = Akense Afasie Test (Graetz et al., 1992), Ant.=anterior, BDAE= Boston Diagnostic Aphasia Examination (Goodglass et al., 2000), BNT= Boston Naming Test (Kaplan et al., 1983), CT= computed tomography, d= day(s), F=female, inf.= inferior, Judgment of line orientation (Benton et al., 1983); m= month(s), M=male; MCA= Middle Cerebral Artery, MMSE= mini mental state examination (Folstein et al., 1975), MRA= Magnetic Resonance Angiography, MRI= Magnetic Resonance Imaging, NI=not indicated, L= left, post.= posterior, R= right, R*=right-handed, ROF= Rey-Osterrieth Figure (Rey, 1941), RCPM= Raven Colored Progressive Matrices (Raven, 1976; Raven & Court, 1998), SPECT= Single Photon Emission Computed Tomography, sup.= superior, Visual form Discrimination task (Benton et al., 1983), w= week(s); WAIS-NL= Wechsler Adult Intelligence Scale, Dutch version (Kooij et al., 2004), WCST= Wisconsin Card Sorting test (Grant et al., 1993), WMS-R= Wechsler Memory Scale-Revised (Wechsler, 1987), y= year(s).

Case	**Age/ gender/ handedness**	**Etiology**	**Lesion localization**
Case 1: Hwang et al. (2001)	40/F/R* monolingual	Infarction	MRI at onset: normal SPECT on day 1: focal hypoperfusion defect L lateral temporal region and crossed cerebellar diaschisis MRI at 2y: normal SPECT at 2y: normal
Case 2: Verhoeven & Mariën (2002)	53/F/R* polyglot	Infarction	CT/MRI: L hemisphere lesion involving inf. front. gyrus, precentral gyrus, ant. insula, postcentral gyrus and supramarginal gyrus SPECT 33d. post-stroke: L frontoparietal hypoperfusion extending to L temporal and adjacent subcortical regions, crossed cerebellar diaschisis SPECT after 3 years: unaltered supratentorial perfusion pattern, improved perfusion in the R cerebellum

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Speech/language characteristics	**Original accent**	**Newly acquired accent**	**Cognition**
No aphasia, dysarthria or AoS but minimal decrease of fluency (by self report), tone errors, FAS Within 3d: almost complete remission of FAS; At 2y : occasional problems with tone in daily conversations	Mandarin	American English-like accent	No memory loss, digit span: normal
At onset: verbal mutism for 1 day evolving to AoS and FAS (no aphasia); 11d – 27d post-onset: AoS and FAS (no dysarthria or aphasia) 36m post-onset: remission FAS, mild residual AoS	Dutch	French, Russian-like	1m post-onset: MMSE, RCPM, WAIS-NL, Hierarchic Dementia Scale, WMS-R, ROF, WCST, Stroop test, Right-Left Orientation test, Visual Form Discrimination test, Judgment of Line Orientation test: above average cognitive

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Case	Age/ gender/ handedness	Etiology	Lesion localization
Case 3: Mariën & Verhoeven (2007) (case 2)	61/M/R* formal test	Hemorrhage	CT: L hemisphere hemorrhage involving the putamen, genu and post. limb of internal capsule with extension to the post. insula, the medial temporal lobe and the paraventricular white matter of the parietal lobe SPECT: 1m post-onset: severe hypoperfusion L thalamus, lentiform nucleus, L medial and lateral temporal region and L motor cortex, crossed cerebellar diaschisis; 6m post-onset: remission R cerebellar hypoperfusion, unchanged perfusion deficits at the supratentorial level

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Speech/language characteristics	Original accent	Newly acquired accent	Cognition
			results (IQ, memory, language, praxis, gnosis, concentration, problem solving, executive functions)
Onset: verbal mutism, global aphasia After 6d: conduction-like aphasia and FAS 1m post-onset: conduction aphasia and agrammatism, FAS, very high speech rate, omissions and consonant deletions, harsh voice quality, syllable-timed and isochronous speech rhythm, groping AAT: weak oral and written comprehension, very weak repetition, esp. words and sentences; weak naming, weak written language 6m post-onset: near remission of auditory-verbal and written comprehension deficits, normal naming, residual conduction aphasia, improved speech rate, improved groping and speech rhythm, FAS receded AAT: improvement comprehension skills, repetition still weak, naming stabilized, only sentences remained weak; written language stabilized) and BNT: 14/60 (-10.63 SD!) (>< AAT naming - total: -1.46 SD) verbal fluency: weak	Dutch	North-African	1m post-onset: MMSE (23/30), RPM (IQ: 117), ROF, Right-Left orientation task, Visual Form Discrimination task, Judgment of Line Orientation: all normal

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C. APPENDICES TO CHAPTER 5

Appendix C1. Overview of the psychogenic case reports (literature review: 1907- July 2014). Relevant information (from left to right) includes the age, gender and handedness of the patients, their medical history, the neurological and neuroradiological exams, the psychological or psychiatric affectation, the accent, and the comorbid speech and language disorders.

Case	**Age/ Gender/ Handedness**	**Medical history**
[1] Critchley (1964) (Case 1) = Critchley (1970 – Case 1)	49/F/NI	/
[2] Gurd et al. (2001)	47/F/R	/
[3] Reeves & Norton (2001) = case 3 Reeves et al. (2007)	65/M/NI	Psychotic exacerbations since thirties, schizophrenia at forty, Parkinson's disease with tremor in bilateral upper extremities, hypertension
[4] Van Borsel et al. (2005)	32/F/NI	Permanent right-sided neurosensory hearing loss with sloping configuration (as of the age of 6); age 23: head trauma and whiplash injury → chronic headache; Age 32: minor head trauma→ hoarseness → ENT exam was normal onset of speech problems shortly of visit to ORL; on-going psychiatric history: depression (suicidal ideation); family problems.

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Neurological, biological, physical and/or radiological examination(s)	**Psychological/ psychiatric affectation**	**Accent**	**Comorbid speech and language disorders/ symptoms**
/	Post-traumatic neurosis after head injury	English Welsh	/
Nov. 1999: normal Doppler, normal MRI, CT: small high signal lesion in cerebellar vermis; Dec. 1999: tone, power, coordination, and reflexes in arms and legs were normal, gait disorder; MRI: several small foci of T2 hyperintensity in peripheral white matter of both frontal lobes, left inf. frontal corona radiata and left thalamus, EEG: sharp and slow waves, but no history of epilepsy; presence of oligoclonal bands in CSF	MS (?)	English (North Yorkshire) French	/
MRI scan (with contrast): normal, Blood and histological exam: normal	(Positive) schizophrenia	American English British English	/
No motor or sensory abnormalities; coordination, gait and posture: normal; CT: normal	Psychological impact, family problems + suicidal ideation	Dutch 'awkward' accent	Mute (initially), agrammatism

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Case	Age/ Gender/ Handedness	Medical history
[5] Verhoeven et al. (2005) = Verhoeven et al. (2013, case 3)	51/F/R formal test, polyglot	Disrupted speech and gait problems since 1995; wheelchair-bound; no history of developmental or psychiatric disorders
[6] Reeves et al. (2007), Case 1	30/M/NI	10-year history of schizophrenia,
[7] Reeves et al. (2007), Case 2	53/F/NI	30-year history of bipolar disorder
[8] Poulin et al. (2007) = Roy et al. (2012)	74/M/R	Epilepsy between 6-14 years; Bipolar disease as of 1982, multiple exacerbations; FAS first mentioned in 2003; Delirium due to lithium intoxication 6 months before FAS started; tremor; neurosensory hypoacusia

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Neurological, biological, physical and/or radiological examination(s)	Psychological/ psychiatric affectation	Accent	Comorbid speech and language disorders/ symptoms
Two months after 'near-accident' (1995): CT: normal; EEG: normal Repeat investigation in 2003: Gait: unsteady, wide-based, coordination, muscle tone and tendon reflexes: normal; CT and (struct.); MRI: normal; EEG: normal; laboratory studies, lumbar punction: normal	Psychotrauma → conversion disorder 1995: conversion disorder (MMPI) 2003: DIS-Q & MMPI: near normal	Dutch (The Netherlands) → French	Paragram-matism?
Laboratory work-up, physical examination: normal; MRI scan: normal; EEG: normal; Blood exam: normal; SPECT: normal	Positive schizophrenia	Southern American English accent → Jamaican accent	/
Laboratory work-up, physical examination: all normal; MRI scan: normal; EEG: normal; Blood exam: normal; SPECT: normal	Psychosis (bipolar disorder)	American English → 'European'	/
Neurological examination: Coordination and gait: decomposition of the half-turn, slight incoordination of left arm, micrographia; Primitive reflexes: palmomental and snout reflexes present; Radiological examination: MRI (Dec. 2005): normal, although slight atrophy in left sylvian fissure; 18-FDG PET scan: diffuse hypometabolism in frontal, parietal and temporal lobes and focal deficit concerning esp. the left sylvian sulcus	Bipolar disorder; recurrent psychotic episodes with manic exacerbations	Québec French → Acadian French/French of France/ English	Mild agrammatism (as of 2002/2003), surface agraphia, Spanish and German-sounding words come to mind: not able to suppress

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Case	Age/ Gender/ Handedness	Medical history
[9] Tsuruga et al. (2008)	44/F/NI	End-thirties: nausea, vomiting, diarrhea, tinnitus, tired eyes, irritations, diagnosed with autonomic imbalance; Few years later: respiratory paroxysm, experienced aphonia (few hours) (hospitalized several times), after violent familial experience: aphonia (2 days), loss of appetite Later: FAS
[10] Haley et al. (2010)	36/F/NI polyglot: late bilingual (Spanish)	Admission: gait: unsteady; posture: left-sided weakness, sensory: visual blurring, altered hearing left ear, slurred speech, weakness of left side of the face, subtle weakness of left arm and leg. 10 days after symptom onset: speech impairment, trouble swallowing and abnormal sensations in the left face, arm, and leg. 5 days later (stroke specialist): symptoms worsened, FAS was diagnosed
[11] Cottingham & Boone (2010)	36/F/R	Several hospitalizations for symptoms not explicable by neurological cause (e.g. sudden hoarseness of voice)

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Neurological, biological, physical and/or radiological examination(s)	Psychological/ psychiatric affectation	Accent	Comorbid speech and language disorders/ symptoms
Laboratory work-up: liver and thyroid: mild, although undefined abnormalities; MRI, SPECT, and EEG: normal	Conversion Disorder	Japanese → Chinese	/
Acute: MRI: normal echocardiogram: moderate mitral regurgitation (also two years prior), Blood analysis: normal. Impression of Bell's palsy, with additional conversion disorder symptoms. Follow-up**:** MRI (10 days later): no abnormalities, MR angiogram: no abnormality of the brain vasculature, CSF: no MS Over subsequent months**:** several relapses, periods with less accented speech, another brain MRI and cervical MRI during relapse: normal	Conversion disorder	English → French, Spanish, Jamaican, Caribbean, African	/
Motor vehicle accident CT: (head): normal Headaches 3 days after accident, facial numbness, weakness in right arm, speech difficulties: 10 days after accident. Later: deafness to left ear Approx. 10 days post-onset: EEG, brain MRA, MRI: normal, neurological examination: normal, but: AoS + left-sided give-way weakness (non-neurological sign), dysarthria	Minor TBI/ Conversion Disorder (?)	English → Eastern European accent (3 years after accident)	Initially dysarthric- or speech apraxic-like symptoms, telegraphic speech

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Case	Age/ Gender/ Handedness	Medical history
[12] Jones et al. (2011)	39/F/R	Unremarkable
[13] Lewis et al. (2013)	54/F/NI	Unremarkable
[14] Polak et al. (2013) case 1	47/M/NI	Refractory OCD for over 25 years
[15] Polak et al. (2013) case 2	65/M/NI	Refractory OCD for over 50 years

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Neurological, biological, physical and/or radiological examination(s)	Psychological/ psychiatric affectation	Accent	Comorbid speech and language disorders/ symptoms
One month after symptom onset: sensory loss, effort-dependent inconsistencies in strength when testing extremities, gait: disturbed, fluctuations, uneconomic postures, dramatic give way weakness; positive 'chair test'; speech: disrupted articulation and prosody; CT, MRI (brain + cervical), EEG: normal	Conversion Disorder	American English Jamaican accent	Initially mute
CT (brain): normal	Mania	American English → Caribbean English	/
March 2006: 2 DBS electrodes → treatment Pre-operative MRI and post-operative CT: no lesions	Refractory OCD (for > 25 years)	Standard Dutch → Pronounced regional Dutch accent	/
/	Refractory OCD	Regional Dutch variant → more sophisticated/ formal Dutch	/

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D. APPENDICES TO CHAPTER 7

Appendix D1. Neuropsychological test results for the years 2012 and 2014. FSIQ= full scale IQ, WAIS-IV= Wechsler Adult Intelligence Scale - IV, WMS-R= Wechsler Memory Scale, Stroop= Stroop task.

NEUROPSYCHOLOGY		
TESTS 2012	**SCALED SCORES**	**MEAN (+- 1SD)** ***Z-score (pct.)***
Intelligence		
WAIS-IV		
FSIQ	105	100 (+-15)
Verbal Comprehension Scale	96	100 (+-15)
- similarities	(10)	10 (+-3)
- vocabulary	(9)	10 (+-3)
- information	(9)	10 (+-3)
Working Memory Scale	112	100 (+-15)
- arithmetic	(14)	10 (+-3)
- digit span	(10)	10 (+-3)
Perceptual Organization Scale	120	100 (+-15)
- block design	(13)	10 (+-3)
- matrix reasoning	(14)	10 (+-3)
- picture completion	(13)	10 (+-3)
Processing Speed Scale	86	100 (+-15)
- symbols	(7)	10 (+-3)
- coding	(8)	10 (+-3)
Attention		
d2-test		
- Total items (Tn)	(249)	*Z=-3.08*
- Total (corrected for mistakes) (Tn-F)	(246)	*Z=-2.94*
- Variation in tempo (Tn highest-Tn lowest)	(7)	*Z=-1.15*
Barrage de Zazzo (10 min.)		
Fastness	(103.6)	*(pct. 12.5-25)*
Exactness	(11.48)	*(pct. 25-50)*
Profitableness	(239)	*(pct. 25-37.5)*
Executive functions		
Wisconsin Card Sorting Test		
Nr. of categories realized	6	
Learning capacity	24.25%	
Nr. of errors	7 (69)	

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TESTS 2014	SCALED SCORES	MEAN (+- 1SD) *Z-score (pct.)*
Attention		
d2-test		
- Total items (Tn)	(242)	*Z=-2.44*
- Total (corrected for mistakes) (Tn-F)	(242)	*Z=-2.20*
- Concentration (C-F2)	(105)	*Z=-1.68*
- Variation in tempo (Tn highest-Tn lowest)	(10)	*Z= 0.5*
Executive functions		
Trail Making Test		
- Test A	(52″38)	*pct.<10*
- Test B	(1'38″25)	*pct. 20*

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NEUROPSYCHOLOGY			
TESTS 2012	**SCALED SCORES**	**MEAN (+- 1SD)** ***Z-score (pct.)***	
Stroop			
Naming	(89″)	64.78 (+- 16.25)	
- Mistakes	2	1.13 (+- 1.59)	
Reading	(39″)	46.72 (+- 16.4)	
- Mistakes	1	0.38 (+- 0.72)	
Interference	(155.8″)	111 (+- 27.58)	
- Mistakes	3	3.5 (+- 4.15)	
Flexibility	(221″)	133.52 (+-52)	
- Mistakes	6	2.89 (+-2.61)	
(Long Term) Memory			
California Verbal Learning Test			
List A	(66)	57.88 (+-5.46)	
Total 1-5	(5)	7 (+-2.37)	
List B			
Free recall of A	(14)	12.35 (+-1.97)	
Cued recall A	(16)	13 (+-1.90)	
Delayed recall A	(15)	13 (+-1.84)	
Cued delayed recall A	(16)	13.59 (+-1.91)	
Recognition	(16)	14.71 (+-1.40)	

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	TESTS 2014	SCALED SCORES	MEAN (+- 1SD) *Z-score (pct.)*
	Memory		
	WMS-R		
	Attention/ Concentration	**(50) 70**	100 (+-15)
	- mental control	(4)	10 (+-3)
	- number series	(18)	10 (+-3)
	- visual series	(28)	10 (+-3)
	Visual Memory	**(66) 133**	100 (+-15)
	- perceptual memory	(7)	10 (+-3)
	- associated visual pairs	(18)	10 (+-3)
	- visual reproduction	(41)	10 (+-3)
	Verbal Memory	**(42) 74**	100 (+-15)
	- logical memory	(26)	10 (+-3)
	- associated verbal pairs	(16)	10 (+-3)
	Global Memory	**(108) 86**	100(+- 15)
	Delayed Recall	**(74) 91**	100 (+-15)
	- logical memory	(11)	10 (+-3)
	- associated visual pairs	(12)	10 (+-3)
	- associated verbal pairs	(14)	10 (+-3)
	- visual reproduction	(16)	10 (+-3)
	Language		
	Boston Naming Test (/60)	(53)	

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E. APPENDICES TO CHAPTER 8

Appendix E1. Transcription oral picture description (November 2014).

PATIENT:

"Der is ene mijnheer die is [eh] ziek [eh] is. Hij heeft ene wein auf die tefel stehen mit eine glas und er zijn boeken auf die tefel ein juffrouw [eh] zit aan hem. Zij... hij... zij trekken aan zijn jas, zij doet wijzen naar die [ehm] radio... en er is een kleine baby op die grond bij een beer en daar is ook [eh] die kat [ehm] op die radio [eh] op de plank. Hij doet vissen [eh] nemen uit die [eh] kom en alle boeken en vasen is op mijnheer komen... op zijn hoofd en daar is een pan koken daar [eh] hierzo een pan maken... koken ... CD maken hier ... en ja ..."

TRANSLATION:

"There is a sir who is [erhm] sick [erhm] is. He has a wine standing on the table with a glass and there are books on the table a miss [...] touches him. She ... he... she pull his jacket, she does pointing to the [erhm] radio... and there is a little baby on the floor near a bear and there also is [erhm] the cat [erhm] on the radio [erhm] on the shelve. He is doing fishing [eh] taking from the [...] bowl and all the books and vases come on the sir ... on his head and there is a pan cook there [egh] here a pan make... cook... CD make here... and yes..."

Appendix E2. Reading text presented to the patient during the neuropsychological assessment in November 2014.

Er worden in de hele wereld veel verschillende talen gesproken. Niemand weet hoeveel talen er precies bestaan. Waar de talen vandaan komen, kan als volgt verklaard worden: in de Bijbel staat het verhaal van de toren van Babel. De mensen wilden een toren bouwen tot in de hemel, maar God strafte hen. Hij zorgde ervoor dat iedereen opeens verschillende talen sprak. Zo konden de mensen elkaar niet begrijpen en het werk aan de toren liep daardoor in het honderd. Waarschijnlijk is het in werkelijkheid anders gegaan. De eerste mensen op aarde spraken één taal. Omdat er niet voldoende eten was voor iedereen, trokken kleine groepjes mensen weg, die groepjes vestigden zich op een andere plaats. Geleidelijk aan gingen ze andere woorden gebruiken. Sommige woorden die ze in hun vroegere omgeving gebruikten, hadden ze in hun nieuwe woongebied niet meer nodig. Voor zaken die ze niet kenden, moesten ze nieuwe benamingen uitvinden. Ook de uitspraak en de manier waarop ze zinnen bouwden, veranderden. Naar verloop van tijd verschilde de manier van spreken zo sterk van de oorspronkelijke taal dat het een nieuwe taal geworden was. Soms kwam een groepje in contact met een groep mensen die een andere taal sprak, die twee talen vermengden zich en zo ontstond een

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nieuwe taal. Uit de ene oorspronkelijke taal hebben zich verschillende taalfamilies ontwikkeld en uit die verschillende taalfamilies zijn de talen van tegenwoordig ontstaan.

There are many languages that are spoken all over the planet. Nobody knows how many languages exist. Where the languages originate from, can be explained as follows: in the Bible there is story of the tower of Babel. The people wanted to build a tower as high as the sky, but God punished them. He made sure that everybody suddenly started speaking different languages. In that way people couldn't understand each other and the works on the tower became disorganized. It probably turned out differently in reality. The first people on earth spoke one language. Because there wasn't enough food for everyone, small groups of people went away, those small groups sometimes installed themselves at another location. Gradually they started using different words. Some words they were using in their prior environment, they no longer needed in their new living environment. Things they did not yet have a name for, they needed to invent names. The pronunciation and way in which they constructed sentences, also changed. After some time, the way of speaking differed so strongly from the original language that it became a new language. Sometimes a small group came into contact with a group of people that spoke a different language, those two languages blended and in that way a new language developed. Out of this one original language different language families were developed and out of the different language families the languages of today were developed.

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SUMMARY

Foreign Accent Syndrome or FAS is a motor speech disorder that causes patients to start speaking their mother tongue with an accent that is perceived as non-native. The impression of a 'foreign' accent is the consequence of segmental and suprasegmental changes occurring in the maternal language. FAS is usually the result of brain damage (often a stroke). However, in recent years, there has been an increasing amount of reports on FAS in association with psychiatric disorders, or developmental disorders. In this dissertation, we have tried to establish an in-depth neurolinguistic analysis of three different variants of FAS: neurogenic FAS, developmental FAS and psychogenic FAS.

In **Chapter 1**, we gave a historical overview of FAS from its first attestation by Pierre Marie (1907) to the formulation of the operational definition and the FAS criteria by Harry Whitaker (1982). Subsequently, we introduced the taxonomic distinction developed by Verhoeven and Mariën (2010), who described the four FAS variants: neurogenic, developmental, psychogenic and mixed FAS. This dissociation served as a common thread for the layout of the current dissertation. Afterwards, we presented an overview of the linguistic, cognitive, and computational explanations for the mechanisms inducing FAS. These theories were the main ideas against which the results in the subsequent chapters were evaluated. We ended this chapter with an overview of the research objectives of this dissertation.

In **Chapter 2** we focused on the neurogenic FAS variant. The chapter constituted a detailed review of the vascular FAS cases published between 1907 and October 2016. We focused on the vascular FAS cases, because these provide the best lesion-behavior correlations. We investigated the demographic data (gender, linguistic background etc.), lesion location , linguistic characteristics, associated cognitive impairments, comorbid speech- and/or language disorders, and onset and remission. We concluded that vascular FAS affects mainly monolingual women, who usually suffered structural damage affecting the premotor cortex, the motor cortex, the insula or the basal ganglia. In terms of the linguistic characteristics there was a preponderance of vowel elongation, vowel fronting, raising, backing and lowering, consonant substitution and consonant omission. On the suprasegmental level, speech was characterized by a slow speech rate, altered rhythm and intonation. The results also showed that vascular FAS is infrequently associated with cognitive deficits, although it often co-occurs with other speech- and language disorders (primarily aphasia). The onset of FAS is usually acute (occurring within 3 weeks after a stroke). Conclusions with respect to prognosis could not be made as the remission data were only irregularly provided. A comparison with ataxic dysarthria and apraxia of speech demonstrated that vascular FAS shares an important amount of perceptual characteristics with these motor speech disorders. After evaluation of the pathophysiology in conjunction with the clinically attested characteristics it is concluded in this chapter that vascular FAS most likely is a dual component disorder. The disruption in connectivity between areas associated with planning and/or execu-

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tion of speech, usually affecting the cortico-striato-pallidal-thalamic pathways, can explain the clinical phenotype of vascular FAS.

In **Chapter 3** we presented two new cases of foreign accent syndrome after posterior fossa lesions. The speech characteristics of both patients showed resemblances with apraxia of speech and ataxic dysarthria. The speech disturbances and cognitive deficits were explained in light of a disruption in the cerebellar connections with (pre-)frontal areas, and the hypothesis is raised whether FAS can be the result of consequent disruptions in feedforward and feedback motor speech control (in which the cerebellum plays a mediating role). An alternative hypothesis is that maladaptive compensation by unaffected brain areas, for instance the contralateral cerebellum (in case 2), plays a role.

In **Chapter 4,** we described the case of a 17-year-old Belgian boy with developmental FAS and comorbid developmental apraxia of speech (DAS). The neuropsychological and neurolinguistic investigations demonstrated that the patient had a normal IQ, but impaired planning and graphomotor skills (visuo-constructional dyspraxia), set-shifting and goal-oriented cognitive strategies. The patient underwent a Tc-99m-ECD SPECT scan. This revealed a significant hypoperfusion in the prefrontal and medial frontal regions, and in the lateral temporal regions. The hypoperfusion in the right cerebellum almost reached significance. We argued that the linguistic deficits (FAS and DAS) as well as the isolated visuo-constructional dyspraxia can be interpreted as the functional expression of the retained hypoperfusions, affecting areas involved in linguistic and non-linguistic tasks involving planning (speech, but also graphomotor plans for writing), attention and execution. The deficits in combination with the SPECT scan again underline the importance of intact cerebro-cerebellar functional communication.

In **Chapter 5** we reviewed all of the assumed psychogenic FAS cases published between 1907 and July 2014. The cases were analyzed for largely the same aspects as the vascular FAS patients, i.e. demographic data, etiologies, FAS characteristics (segmental and suprasegmental), associated cognitive impairments, comorbid speech- and/or language disorders and onset and remission data. The results demonstrated that psychogenic FAS predominantly affects monolingual women, that the disorder often co-occurs with a mental disorder and that FAS onset is usually associated with exacerbations in the context of an associated psychopathology. In cases in which the patient did not believe the FAS to be of psychogenic origin, the onset of the FAS was often considered particularly late compared to the incident the patient him-/herself held responsible for inciting the FAS (see also **Chapter 7**). The associated linguistic characteristics were primarily vowel elongations, additions, and fronting, and at the consonant level: omissions and substitutions. At the suprasegmental level, speech was marked by an abnormal intonation, a slow speech rate and an altered speech rhythm. The psychogenic patients showed a diverse set of cognitive impairments affecting memory, attention, executive functions, processing speed, intelligence, fine motor skills and visuospatial skills. These impairments were often directly associated with the psychopathology. In case of doubt, the hypothesis of cogniform disorder – a cognitive disorder

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of a somatoform nature – was raised. Comorbid speech and/or language disorders were very specific. Patients sometimes went through a period of mutism and developed grammatical disorders. Symptom evolution was different from the one found in vascular FAS: speech symptoms fluctuated frequently over time, sometimes in association with psychiatric exacerbations. Lastly, we found that psychogenic FAS usually recedes simultaneously with the resolution of the associated psychiatric disorder (often pharmacologically induced), or spontaneously.

In **Chapter 6** we discussed the case of a 40-year-old, non-aphasic, polyglot Belgian woman with a history of drug addiction and psychiatric problems. In February 2010, she acutely developed a foreign accent of sudden onset in her first language (French). Speech was relatively fluent, although the patient sometimes spoke in a stutter-like manner and she had a marked grammatical output disorder. The initial psychodiagnostic assessment disclosed indications for a conversion disorder, although the diagnosis could not be formally objectified. After three years, the patient's accent suddenly receded after a general anesthesia for an appendectomy. Her grammatical output disorder however, remained present, although only in the writing of her first language (French). We conducted a perceptual accent rating experiment in which the raters were native speakers of French. They confirmed that the patient sounded foreign. Based on the irregularities within the neurolinguistic profile, the psychiatric traits, and the sudden spontaneous recovery affecting only one modality, we concluded that the patient suffered a FAS of psychogenic origin.

In **Chapter 7** we presented the case of a 33-year-old, French-speaking Belgian woman who was involved in a car accident as a pedestrian. She developed a German/Flemish-like accent approximately six months *after* the incident. Based on the neuroimaging data in the acute phase, structural brain damage was formally excluded. The neuropsychological investigations revealed poor executive functions, processing speed, attention and verbal memory. We explained these deficits in the context of a post-concussion syndrome. We did not formally rule out cogniform deficits: the fact that the patient was implicated in litigation when she consulted the department, warranted such a hypothesis. Importantly, the accent – which fluctuated over time – was first noticed when the patient was dismissed from work. The psychodiagnostic evaluation disclosed that there were indications for a borderline personality although a formal diagnosis could not be made. We equally performed a perceptual accent rating experiment same methodology as in chapter 6 – which confirmed that the patient was perceived as a non-native speaker of French by a group of native French-speaking raters. Specifically, the symptom evolution, including delayed onset and psychodiagnostic findings in the absence of neurological disorder, argued in favor of a diagnosis of psychogenic FAS.

In **Chapter 8**, we discuss the case of a 28-year-old Dutch-speaking woman from the Netherlands who developed FAS two weeks after she slipped and fell down the staircase. Medical history revealed that around the same time, the patient had started displaying very disoriented behavior and had visual hallucinations. The FAS was unlike

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any previously published case: when in stress, the accented speech was interspersed with German(-like) words and structures (language mixing and switching) and sometimes evolved to neologistic jargon speech. Apart from a borderline low intelligence, the formal psychodiagnostic tests did not lead to the diagnosis of a psychiatric disorder. The contradictory findings in the neurolinguistic tableau, in conjunction with her psychiatric and speech symptoms, were strongly indicative of an underlying psychogenic etiology.

In **Chapter 9** we discussed the main research findings of this dissertation. In this final chapter we compare the three types of FAS that were described and discussed in this dissertation. The status of FAS as a motor speech disorder at the level of execution and planning is evaluated as well as the role of the cerebellum in its pathophysiology. We end the chapter concluding that the term 'foreign accent syndrome', as coined by Whitaker, is a misnomer.

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SAMENVATTING

Foreign Accent Syndrome of FAS is een motorische spraakstoornis die ertoe leidt dat de patiënt zijn/haar moedertaal gaat spreken met een accent dat gepercipieerd wordt als dat van een buitenlander. De indruk van het "buitenlands" accent is het gevolg van veranderingen die optreden in de moedertaal op het niveau van de fonemen en de prosodie. FAS komt meestal voor na hersenschade (vaak een beroerte). Er wordt ook steeds vaker melding gemaakt van FAS na psychiatrische stoornissen of FAS als een ontwikkelingsstoornis. In dit onderzoek hebben we getracht een zo volledig als mogelijke analyse te realiseren van de drie varianten van FAS: neurogene FAS, psychogene FAS en FAS op ontwikkelingsbasis.

In **Hoofdstuk 1** hebben we een uitgebreid historisch overzicht van FAS gegeven, vanaf de eerste vaststelling door Pierre Marie (1907) tot de beschrijving van de FAS-criteria door Harry Whitaker (1982). Vervolgens hebben we het taxonomisch onderscheid geïntroduceerd zoals voorgesteld door Verhoeven en Mariën (2010a). Zij beschreven eerder de vier varianten van FAS: neurogene, psychogene, en gemengde FAS en FAS op ontwikkelingsbasis. Deze onderverdeling heeft de leidraad gevormd voor de verdere opdeling van deze thesis. Hierna hebben we een overzicht gegeven van de linguïstische en cognitieve theorieën en de theorieën uit de computerlinguïstiek omtrent het ontstaan van FAS. Deze hebben het theoretisch kader gevormd dat van belang was bij de interpretatie van de resultaten van de verschillende hoofdstukken. Tot slot hebben we in dit hoofdstuk ook de doelstellingen van dit onderzoek vermeld.

In **Hoofdstuk 2** hebben we de focus op neurogene FAS gelegd. We hebben hierbij de nadruk gelegd op FAS na een beroerte omdat dit de beste correlaties tussen schade en gedrag toelaat. We hebben de invloed onderzocht van demografische gegevens (geslacht, linguïstische achtergrond etc.), lesielokalisatie, linguïstische karakteristieken, geassocieerde cognitieve stoornissen, comorbide spraak- en taalstoornissen, en aanvang en herstel van FAS. In de resultatensectie hebben we geconcludeerd dat FAS na een beroerte voornamelijk eentalige vrouwen treft, die gewoonlijk structurele hersenschade hebben ter hoogte van de premotorische cortex, motorische cortex, insula en/of basale ganglia. Met betrekking tot de linguïstische karakteristieken hebben we een overwicht van klinkerverlengingen gevonden. De klinkeruitspraak in FAS wordt verder gekenmerkt door *fronting*, *backing*, *raising*, en *lowering*. Verder hebben we voornamelijk consonantsubstituties en omissies weerhouden. Op het niveau van de prosodie wordt de spraak gekarakteriseerd door een lage spreeksnelheid, veranderend ritme en aangepaste intonatie. FAS na een beroerte gaat slechts zelden gepaard met cognitieve stoornissen. Anderzijds hebben we aangetoond dat er vaak bijkomende spraak- en taalstoornissen optreden, voornamelijk afasie. De aanvang van FAS blijkt meestal acuut te zijn (binnen drie weken na de beroerte). Conclusies met betrekking tot herstel hebben we niet geformuleerd, aangezien de gegevens omtrent remissie niet consequent

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gerapporteerd worden. Een vergelijking met atactische dysartrie en de spraakapraxie heeft aangetoond dat FAS na een beroerte een groot aantal semiologische gelijkenissen vertoont met deze motorische spraakstoornissen. Na evaluatie van de pathofysiologie en de klinische karakteristieken hebben we in dit hoofdstuk geconcludeerd dat FAS na een beroerte hoogstwaarschijnlijk een stoornis van duale aard behelst. De verstoring in de connecties tussen gebieden die geassocieerd zijn met planning en/of executie van de spraak – en die vaak cortico-striato-pallidale-thalamische verbindingen betreffen – kunnen het klinisch fenotype van deze stoornis verklaren.

In **Hoofdstuk 3** hebben we twee gevallen van FAS na schade in de posterieure fossa beschreven. Na vaststelling van de spraakkarakteristieken van beide patiënten hebben we duidelijke gelijkenissen gevonden met het klinisch beeld voor de atactische dysartrie en spraakapraxie. De spraakkarakteristieken en de bijkomende cognitieve stoornissen hebben we verklaard in het kader van een verstoring in de cerebellaire connecties met (pre-)frontale regio's. We hebben hier de hypothese gesteld dat FAS het gevolg kan zijn een verstoring van de feedforward- en feedbackcontrolemechanismen in de spraakproductie. We hebben ook een alternatieve hypothese geformuleerd, namelijk dat een intact hersengebied, misschien de andere cerebellaire hemisfeer (bijvoorbeeld in de tweede patiënt), mogelijk een abnormale compensatie heeft bestendigd.

In **Hoofdstuk 4** hebben we de gevalstudie beschreven van een 17-jarige Belgische jongen met FAS op ontwikkelingsbasis en een verbale apraxie, een normaal IQ, maar sterk verstoorde planningsfuncties (visuoconstructieve dyspraxie). Een SPECT scan heeft gewezen op een significante hypoperfusie in de prefrontale en mediaal frontale regio's en in de lateraal temporale regio's. We hebben in dit hoofdstuk geargumenteerd dat de linguïstische problemen (FAS en verbale dyspraxie) en de geïsoleerde visuoconstructieve apraxie een functionele expressie zijn van de vastgestelde hypoperfusies. We hebben in dit hoofdstuk getracht het belang van een intacte functionele cerebro-cerebellaire communicatie te onderstrepen.

In **Hoofdstuk 5** hebben we een kritisch overzicht van alle veronderstelde psychogene casussen opgesteld (gepubliceerd tussen 1907 en juli 2014). We hebben de gevalstudies geanalyseerd voor dezelfde aspecten als bij FAS na een beroerte (hoofdstuk 2); namelijk de demografische gegevens, etiologieën, FAS karakteristieken (op foneemniveau en op niveau van de prosodie), geassocieerde cognitieve stoornissen, comorbide spraak- en/of taalstoornissen, en aanvang en herstel. We hebben aangetoond dat psychogene FAS voornamelijk eentalige vrouwen treft en verder dat de stoornis vaak voorkomt met psychiatrische stoornissen. De aanvang van FAS wordt vaak geassocieerd met een toename van positieve psychiatrische symptomen (bijv. psychose bij schizofrenie). Met betrekking tot de linguïstische karakteristieken hebben we voornamelijk klinkerverlengingen en toevoegingen van klinkers weerhouden, maar ook het meer naar voren uitspreken van deze klanken (ook wel *fronting* genoemd). Op het consonant-

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niveau hebben we voornamelijk omissies en substituties vastgesteld. Met betrekking tot de prosodie hebben we een abnormale intonatie, een traag spreektempo en een aangepast ritme als meest kenmerkend geduid. Psychogene patiënten vertonen verscheidene cognitieve stoornissen die geheugen, aandacht, executieve functies, verwerkingssnelheid, intelligentie en visuospatiële capaciteiten betreffen. De comorbide spraak- en/of taalstoornissen zijn – als ze voorkomen – vaak zeer specifiek. De analyse heeft uitgewezen dat de patiënten soms een periode van mutisme doormaken en in sommige gevallen ook grammaticale stoornissen ontwikkelen. We hebben vastgesteld dat de evolutie van de symptomen anders is dan voor de patiënten die FAS ontwikkelen na een beroerte. Het onderzoek heeft bijvoorbeeld aangetoond dat symptomen door de tijd heen kunnen fluctueren en soms samen met de psychiatrische klachten toenemen. Tot slot hebben we geconcludeerd dat herstel meestal het gevolg is van farmacologische onderdrukking van de psychiatrische klachten. In andere gevallen treedt herstel meestal spontaan op.

In **Hoofdstuk 6** hebben we de gevalstudie van een 40-jarige, polyglotte vrouw met een drugsverleden en psychiatrische problemen besproken. In februari 2010, ontwikkelde ze een buitenlands accent in haar moedertaal, Frans. Haar spraak was vloeiend, hoewel de patiënt soms ook stotterde en een grammaticale productiestoornis vertoonde. Een eerder uitgevoerd psychodiagnostisch onderzoek gaf al indicaties voor een conversiestoornis maar de diagnose kon nooit formeel worden bevestigd. Na drie jaar verdween het accent van de patiënte plots na een algemene anesthesie voor een appendectomie. De grammaticale problemen bleven aanwezig, maar enkel in schrift, in haar moedertaal (Frans). We hebben op basis van spraakstalen van de patiënte een perceptie-experiment uitgevoerd waarbij moedertaalsprekers van het Frans als beoordelaars functioneerden. Zij hebben het accent van de patiënte als buitenlands beoordeeld en aldus de diagnose van FAS bevestigd. Op basis van de eerdere indicaties voor een psychiatrische stoornis, maar ook de fluctuaties binnen de opeenvolgende neurolinguïstische onderzoeken en het spontane herstel dat slechts één modaliteit betrof, hebben we hier geconcludeerd dat deze patiënte een FAS van psychogene oorsprong heeft opgelopen.

In **Hoofdstuk 7** hebben we de gevalstudie van een 33-jarige Belgische vrouw beschreven die betrokken was bij een auto-ongeval als voetganger. Ze ontwikkelde een Duits-/Nederlands-klinkend accent ongeveer zes maanden na dit incident. Medische beeldvorming in de acute fase heeft ertoe geleid dat we structurele hersenschade formeel konden uitsluiten. Na uitgebreid neuropsychologisch onderzoek hebben we bij deze patiënte problemen weerhouden op gebied van executieve functies, verwerkingssnelheid, aandacht en verbaal geheugen. De stoornissen hebben we verklaard in de context van een post-commutioneel syndroom. Toch hebben we cogniforme stoornissen niet formeel uitgesloten: het feit dat de patiënte betrokken was in een rechtszaak tijdens de consultatie rechtvaardigt dergelijke hypothese. Tijdens het eerste onderzoek

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heeft de patiënt erop gewezen dat het accent – dat fluctueerde – voor het eerst werd opgemerkt nadat ze ontslagen werd. We hebben psychodiagnostische evaluaties uitgevoerd die wezen op een borderline persoonlijkheidsstoornis. We hebben ook hier een perceptie-experiment beschreven – uitgevoerd volgens dezelfde methodologie als beschreven in hoofdstuk 6 – en dit heeft geleid tot de bevestiging van de perceptie van het buitenlands accent. We hebben opnieuw de diagnose van een psychogene FAS gesteld op basis van het symptoomverloop, het tijdstip van ontstaan van FAS, de psychodiagnostische gegevens en de afwezigheid van structurele hersenschade.

In **Hoofdstuk 8** hebben we de gevalstudie van een 28-jarige Nederlandse patiënte uitgewerkt die twee weken nadat ze van de trap viel plots FAS ontwikkelde en verward gedrag begon te vertonen (inclusief visuele hallucinaties). Analyse van de spraak van deze patiënte heeft aangetoond dat dit geval van FAS verschillend van alle reeds gepubliceerde gevallen. We hebben in haar spraak niet alleen Duits(-klinkende) woorden en structuren weerhouden ("language mixing" en "language switching"), maar ook een neologistisch jargon. Behalve een laag intelligentiepeil hebben de psychodiagnostische testen geen psychiatrische stoornis aangetoond. Op basis van de contrasterende resultaten op de neurolinguïstische tests, de psychiatrische symptomen en spraaksymptomen hebben we hier geconcludeerd dat de FAS van psychogene oorsprong was.

In **Hoofdstuk 9** hebben we de belangrijkste bevindingen van deze thesis besproken. We hebben de drie varianten van FAS en de status van FAS als een motorische spraakstoornis op het niveau van planning en executie geëvalueerd, evenals de rol van de kleine hersenen of het cerebellum in de pathofysiologie van deze aandoening. We hebben dit hoofdstuk afgesloten met een bespreking van de terminologie, omdat de term "foreign accent syndrome", zoals voorgesteld door Whitaker, niet strookt met de bevindingen.

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Stefanie Keulen was born on March 14th, 1990 in Tongeren, Belgium. In 2012, she obtained a Bachelor in the Arts in Dutch and English Linguistics and Literature from the Vrije Universiteit Brussel in Belgium. In 2013, she graduated from the same institution with a Master in the Arts with a specialization in language pathologies and language psychology. She was awarded the Guillaume Duflou prize for best Master's dissertation in the Germanic languages. In October 2013, she was appointed PhD researcher by Prof. Dr. Peter Mariën (Vrije Universiteit Brussel, BE), Prof. Dr. Roelien Bastiaanse (University of Groningen, NL) and Prof. Dr. Jo Verhoeven (University of Antwerp, BE; and City University London, UK) on a joint research project of the Vrije Universiteit Brussel and University of Groningen on Foreign Accent Syndrome. She enrolled as a doctoral student at both institutions and integrated the Clinical and Experimental Neurolinguistics (CLIEN) research group at the Vrije Universiteit Brussel, and the Center for Language and Cognition (CLCG) at the University of Groningen. She completed a research stay at the University of Groningen for six months during the project.

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